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Bringing up an XY Girl:
Parents' Experience of Having a Child with
Androgen Insensitivity Syndrome.

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ABSTRACT

In androgen insensitivity syndrome (AIS), the masculinising hormones produced by the testes are unable to have an effect on the developing, genetically male (46 XY karyotype) foetus. Children with the condition therefore present with a female phenotype but an absence of uterus and ovaries and, at best, a short, blind-ending vagina. Such children are generally raised as girls (Slijper *et al.*, 2000). Previous research has focused on the experience of the individuals with the intersex condition (e.g. Froukje *et al.*, 1998) but little has been published concerning the impact of the condition on parents. Twenty parents of children with AIS were interviewed and the data analysed using a grounded theory methodology to describe the parental experience.

The first section of the study considers the overall parental experience from the moment of diagnosis. Parents report feeling devastated by the diagnosis and then isolated and uninformed. The central category for the data relating to this section is seen to be one of powerlessness. Parents commonly have concerns about the impact that the disclosure will have upon their children and are unclear about how and when to give the information. The second section of the study focuses on the disclosure issue and the central category here represents the desire to provide protection.

The results of the study offer insight into and enhance understanding of the parental experience. They highlight areas of unmet need which services will need to address and provide many ideas for future research.

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CHAPTER 1 - INTRODUCTION

Androgen insensitivity syndrome (AIS) is a rare intersex condition which causes us to question our traditional views of gender as being distinctly male or female. Parents who have children born with the condition are challenged with the information that though their baby girl appears female on the outside, she is chromosomally male and, rather than female reproductive organs, she has internal testes.

This knowledge is strange and understandably often affects parents deeply. They must come to terms with the information for themselves and then, as their child grows up, they must consider if, how and when to explain about the condition. Research has investigated the biological and medical aspects of this and other intersex conditions and studies have considered the experience of individuals who have become aware of their own diagnosis and the impact that this has had. However, the literature seems largely to have ignored the experience and needs of parents and it is this that the current study seeks to address.

Normal Foetal Development

In the initial development of the human foetus, it is impossible to distinguish male and female reproductive systems and genitalia. Early in development, the Y chromosome possessed by males (who have XY sex chromosomes) causes the neutral gonads to become testes, whereas in females (who have XX sex

chromosomes), the lack of a Y chromosome causes the gonads to develop into ovaries.

In the early weeks of gestation, the genitalia are indistinguishable in male and female foetuses. In both cases there is a midline cleft with a button-like swelling at the top representing the future penis or clitoris. It is not until some twelve to fifteen weeks after conception that, in a male, the testes begin producing a hormone, androgen, and the neutral genitalia begin to develop into a penis and scrotum. Without these androgen levels in the female foetus, the clitoris remains small and the cleft open (Warne, 1997).

Similarly, the internal reproductive organs develop in the growing foetus. In females, the tubes on the back wall of the abdomen (the Müllerian ducts) develop into a female uterus, fallopian tubes and upper part of the vagina. However, in males the testes produce an anti-Müllerian hormone which blocks this process whilst androgen triggers the development of the prostate, seminal vesicles and vasa deferentia from the Wolfian ducts (LeVay, 1993).

What is 'Intersex'?

Broadly speaking, intersexuality constitutes a range of anatomical conditions in which an individual's anatomy mixes typically masculine anatomy with typically feminine anatomy (Dreger, 1998). The cause of intersexuality can be chromosomal or hormonal and intersex conditions vary from children with ambiguous genitalia

who are hard to classify as male or female, to those with much more subtle anomalies (Dreger, 1998).

Classification of Intersex Conditions

The current system of taxonomy for congenital sexual anatomies was devised 125 ago in order to make sense of the confusing anatomies that doctors were presented with and to sort these into clear types. The categorisation hinged on gonadal structure, regardless of the functionality of these gonads or the phenotypic presentation (Dreger, 2000). The system, developed by Klebs (1868), divided humans into five basic types and has largely been adhered to since the taxonomic system was conceived. These five basic types are described briefly below:

- (i) Females - defined as presenting with only standard female sexual anatomy
- (ii) Males - defined as presenting with only standard male sexual anatomy
- (iii) Female Pseudohermaphrodites - defined as presenting some mixture of standard female and male sexual anatomy with the presence of ovaries and an XX karyotype. Generally, individuals with this condition have been exposed to excess androgens in utero causing masculinisation of the external genitalia of a female foetus (i.e. they have ovaries and some aspect of the male genitalia but lack testes).

(iv) Male Pseudohermaphrodites - defined as presenting some mixture of standard female and male sexual anatomy with the presence of testes and an XY karyotype. There is deficient masculinisation of the external genitalia due to problems in the production, synthesis or receptor sensitivity to androgens such that their male genitalia are not sufficiently developed. In these children, the external genitalia are inadequately virilised, the gonads (or testes) are located in the abdomen or inguinal canal, the uterus and ovaries are not in evidence and the vagina has been inadequately formed, if at all. If the insensitivity to androgens is complete, as in complete androgen insensitivity syndrome (CAIS), the child is born with apparently female external genitalia.

(v) True Hermaphrodites - defined as having both ovaries *and* testes or one or more ovotestes. The definition of true hermaphroditism does not depend on the presentation of other sexual anatomy (genital anatomy and reproductive tracts can vary) or a particular karyotype, however, of this group, 80% have the 46XX karyotype (Donahue, 1987).

In recent years, there has been a growing debate about the appropriateness of the above classification system since it does not actually appear to represent a division into 'natural kinds' (Dreger, 2000) and privileges gonadal anatomy regardless of chromosomes, gonadal function or phenotype. Scientists now recognise that gonadal structure does not necessarily correlate with the latter characteristics and that the system we continue to use can cause confusion and psychological harm to individuals with such conditions and their parents (Fausto-Sterling, 1993).

One goal of The North American Task Force on Intersexuality (NAFTI) is to establish a new sex nomenclature to replace the current system with emotionally neutral terminology that emphasises developmental processes rather than perceived gender categories (Fausto-Sterling, 2000).

Androgen Insensitivity Syndrome (AIS)

Androgen insensitivity syndrome (previously known as ‘Testicular Feminisation’) is an X-linked, male pseudo-hermaphroditic intersex condition (Slijper, 2000). A gene alteration results in a defective androgen receptor function such that the masculinising hormones (or androgens - of which testosterone is the leading member) produced by the testes are unable to have an effect on the developing, genetically male (46 XY karyotype) foetus. This means that internally, the male reproductive tract (prostate, seminal vesicles and vasa deferentia) fails to develop. However, there is no development of a female reproductive tract (upper part of the vagina, cervix, uterus, fallopian tubes) and the individual will therefore be infertile (Griffen, 1992).

In terms of physical appearance (or phenotype), children born with complete androgen insensitivity syndrome (CAIS) appear fully female with apparently no masculinisation of the genitals and the only outward signs of the condition being a lack of pubic hair and an absence of menarche at puberty (Broks, 1999). Individuals with partial AIS (PAIS) will have responded, to some degree, to the androgens present prenatally. They are usually born with genitalia which have been

masculinised to an extent and which may appear ambiguous at birth although this can vary from a slightly enlarged clitoris to an almost fully formed penis and scrotum (Warne, 1997).

At birth, children with CAIS are clearly female in appearance and are almost always reared as girls (Slijper, 2000) whereas the sex of a new-born baby with PAIS may be unclear. Some children with PAIS are raised as girls and others as boys, depending on the degree of virilisation (see later discussion about assignment to gender). The children with PAIS whose parents were interviewed for the current study had all been raised as female from birth.

Incidence and Prevalence of Intersex Conditions

It is difficult to estimate the incidence and prevalence of intersexuality. John Money (a psychologist from the John Hopkins University in the USA who was at the forefront of the treatment of intersexuals in the 1960's) placed the frequency of intersexuality as being as many as 4% of live births (reported by Fausto-Sterling, 1993). However, Fausto-Sterling (2000) has since revisited this estimate and now places it at more like 1.7% and Nussbaum (2000) states that intersexuality, in a variety of forms, occurs in about one of every two thousand births (0.2%). However, Blackless *et al.* (2000) believe that 1% of all births will be of a baby whose body differs from the norm in terms of its sexual anatomy and Warne (1997) states that one in every 4500 new-born babies (0.02%) have external genitals that differ.

The exact prevalence of androgen insensitivity syndrome is hard to ascertain with estimates varying from one in every 20,000 'male' births (Broks, 1999) to one in 65,000 of the population (Griffen, 1992) or even one in 99,000 (Slijper *et al.*, 2000). Baron (1994) estimates that among all cases of AIS the frequency of PAIS is 28.3% (roughly a quarter of all cases). Using Slijper *et al.*'s (2000) estimate for the prevalence of AIS, this would mean that complete AIS (CAIS) occurs at a rate of one in 132,000 and partial AIS (PAIS) occurs at the rate of one in 396,000.

The Diagnosis of an Intersex Condition

The physical features that initially suggest the possibility of AIS can differ from individual to individual and these differences may influence the age at which the condition is diagnosed (Warne, 1997). AIS is commonly diagnosed in the first few days, weeks or months after birth when the testes, in an attempt to migrate down into the non-existent scrotum, become lodged in the groin where they can be felt as lumps and are often thought to be inguinal hernias (LeVay, 1993). In operating to repair the hernias, the surgeon may find that the hernias contain testes and further investigations are then necessary.

Where hernias go unnoticed or are not present, it may not be until the time at which puberty is expected that anything is felt to be unusual. Because the testes remain in the body, breast development and the development of a female body shape will, in these cases, be normal due to the conversion of androgen to oestrogen in the body.

However, there will be an absence of menarche (periods) in addition to the lack of development of both pubic and armpit hair.

To confirm the diagnosis of an intersex condition the medical team will usually undertake a blood test to identify the chromosome pattern and a full physical examination of the child including ultrasound, to look for the presence and position of gonads, any genital abnormalities and to look for the presence and position of uterus and vaginal cells. Once XY chromosomes and testes are detected, there may be an assessment of the individual's ability to produce testosterone when stimulated with human chorionic gonadotrophin (hCG). This is important in distinguishing CAIS from other medical conditions where the XY child has testes which are not able to produce androgen.

The Impact of Diagnosis and Parental Adjustment

Although there is relatively little literature which considers the impact on parents of receiving an intersex diagnosis for their child, there are a small number of studies which consider the outcomes for parents receiving a diagnosis of developmental disability for their child. There is also a limited body of literature concerned with parental reactions to the diagnosis of chronic illness in their child. Clearly these conditions are quite different in nature to a diagnosis of AIS but it is likely that there will be some aspects of the experience of receiving the diagnosis that are similar for parents in all of these groups.

Blacher (1984) states that the concept of a stage model of adjustment for parents of children born with 'handicaps' has been present since the mid 1950s (eg Blodgett, 1957) and has persisted such that there are now many descriptions in the literature of discrete, sequential stages of adjustment. The phases described by different authors can be organised into three, broadly chronological areas (Blacher, 1984).

Firstly there is said to be an initial crisis response which includes immediate reactions in the form of shock, denial and disbelief. Authors have described responses in this stage which include detachment, bereavement and bewilderment, (eg Emde and Brown, 1978). Secondly, there is a phase labelled continuing feelings and responses where authors refer to emotional disorganisation which is characterised by feelings of anger, resentment, aggression, disappointment, guilt and lowered self esteem in the parents. It is at this stage where parents may search for reasons for their child's disabilities and blame themselves for the way in which they have been affected (Blacher, 1984). Finally, the third stage is described as the adjustment or acceptance phase and is said to be characterised by emotional organisation, adjustment, acceptance or reorganisation. It is only at this time that parents are said to become less anxious and to grow more comfortable in their situation (Wikler *et al.*, 1981).

Although there is some variation between different authors' accounts of the type and length of the stages of adjustment, on the whole, the literature on parental stages of adjustment appears to be reasonably consistent. However, it has been claimed that research into this stage model is based upon 'popular wisdom' (Blacher, 1984, pp 65)

which has been reinforced and agreed over the years and which may oversimplify an extremely complex process of adjustment. A critical question remains as to whether the number and character of the stages described and agreed exist for parents today, and specifically for those parents of children diagnosed with an intersex condition such as AIS.

Pianta *et al.* (1996) have studied reaction to diagnosis from an attachment theory perspective. Whilst the attachment system is said to utilise child behaviours to maintain proximity to the parent in times of threat, the caregiving system is hypothesised as a complimentary system in the parent which also has the biological function of protecting the child from harm (Bowlby, 1969, George and Solomon, 1996). Important elements of the caregiving system include parental beliefs, attributions, perceptions and expectations for and about their child which can affect a parent's behaviour towards their child (Melson *et al.* 1993). In addition, emotional experience (such as depression or anger) is also said to be a vital component and affects caregivers' responses to the child with the diagnosed condition (Cohn *et al.* 1990).

Pianta *et al.* (1996) hypothesised that learning that one's child has a disability or chronic illness acts as a possible trauma to the caregiving system by introducing information and experience about a child that challenges existing beliefs, feelings and expectations. Using the 'Reaction to Diagnosis Interview' (Pianta and Marvin, 1992a), the authors assessed resolution of the loss or trauma associated with learning of their child's chronic illness or disability and found that whilst all the mothers were

believed to have received similar levels of information and support concerning their child's condition and comparable levels of involvement in their medical care, some 50% of them remained unresolved in respect to the diagnosis. Pianta *et al.* (1996) found a strong link between the parents' resolution of the diagnosis and the quality of their child's attachment to them and stated that the unresolved parents had not yet integrated the experience of the diagnosis within their views of themselves and their relationships.

In investigating the reasons for parental resolution or non-resolution of the diagnosis, Pianta *et al.*, (1996) found that neither the severity of the child's condition, nor the child's developmental age had an impact. Sheeren *et al.*, (1997) further investigated the particular factors that influence maternal resolution of diagnosis and found that resolution was linked to 'social and ecological factors' (Sheeren *et al.*, 1997, pp 199) which included the amount of parenting stress experienced, the level of marital satisfaction and the amount and helpfulness of social support. They concluded that a mother's capacity to successfully grieve the loss of the perfect child is related not only to individual and parent-child adjustment (Pianta *et al.*, 1996) but also to the family and social constructs of parenting stress, marital quality and social support (Sheeren *et al.*, 1997). Relationships and support were seen as creating a context where integrative mental strategies could work towards resolution of the experience (Marvin and Stewart, 1990).

The Experience for Parents of Receiving an Intersex Diagnosis for their Child

A medical diagnosis that affects the genital and reproductive organs is said to be a particular challenge to understand, discuss and come to terms with (Warne, 1997). Slijper *et al.*, (1998) believe that confrontation with an intersex condition can be a traumatic experience for parents. Extensive literature reviews have revealed just one study by Slijper *et al.* (2000) which focuses on the impact on Dutch parents' of receiving the diagnosis. However, there appear to have been no specific studies considering this aspect within the UK or the USA.

Slijper *et al.* (2000) studied the emotional reactions of 33 parents (18 mothers and 15 fathers) to the clinical diagnosis of AIS (both partial and complete) in a quantitative study. Firstly, parents were interviewed concerning their psychological reaction to the clinical diagnosis. Feelings of shock, sadness, guilt and shame at that time were later rated as being either 'present' or 'absent' both at the diagnosis and at the time of the study. They found that the majority of both mothers and fathers reacted with shock, grief, anger and shame, although grief was more commonly recorded for the fathers and the mothers showed stronger feelings of guilt which may have been related to having been a carrier or potential carrier of AIS. As time passed, feelings of grief and shame were seen to remain for both parents, whilst mothers additionally retained feelings of shock, anger and guilt.

Secondly, the interview material was rated on a five point scale from 'not at all' to 'completely' for parental acceptance of various aspects of AIS including (i) the discrepancy between the genotype and phenotype, (ii) the appearance of the external genitalia, (iii) the infertility of the child and (iv) the carrier status of the mother. Slijper and colleagues found that parents of children with CAIS or PAIS reported that they found the discordance between the genotype (XY male) and phenotype (female) of their child one of the most difficult aspects to deal with. However, parents of girls with PAIS (i.e. with some masculinisation of the genitalia at birth) reported finding it hard to cope with what they perceived as cross-gender behaviour in their daughter. The infertility of their child was reported to be difficult for 84% of mothers and 75% of fathers and approximately half of all the parents expressed concerns that their child would have reduced chances of finding a partner.

Thirdly, the study considered the support that parents felt that they received throughout this time (again rated on a five point scale) and a questionnaire measure assessed social support in general. Some 28% of parents felt that AIS had had a negative impact on their relationship with their partner and 40% of the mothers indicated a lack of support from their partner in coping with AIS. Only a third of the parents in the study reporting having received psychological support at the time of diagnosis. Factors which Slijper *et al.* (2000) had felt had been important in determining parental coping included receiving full information about AIS when the clinical diagnosis was made. Additionally, they reported that the time of diagnosis appeared to have some influence on parents' coping, with a diagnosis of AIS after the onset of puberty causing parents to suffer more shock and grief but less shame.

Receiving the diagnosis at puberty appeared to give parents less time in which to prepare the news compared to parents who are informed about AIS in their child's early life.

Finally, questions were asked concerning how parents had informed their children of the diagnosis of AIS. The interviews were supported with additional questionnaire measures to assess the impact of the diagnosis of AIS on parents as well as any feelings of hopelessness for the future or depression. Fathers and mothers were then compared in their responses statistically, using chi-square tests. However, the study gives very little detail on this aspect. They report simply that eight of the parents (four couples) received the diagnosis and subsequent information in the presence of their daughters, that another two children (out of 20) received complete information from their parents, that one received the information from her gynaecologist and that those who had not been told of their diagnosis were prepubertal.

Issues of Gender in Intersex

Children with AIS are raised as female yet have male sex chromosomes and are born with internal testes. This information challenges traditionally held views of gender and the Slijper *et al.* (2000) paper notes that parents may find this contradiction hard to deal with. It is important to consider, therefore, what is known about the development of various aspects of gender for children with intersex conditions and the role that both biology and the environment might play in this.

In general 'sex' has been used to refer to the biological categories of 'male' and 'female' and is categorised by chromosomes and genitalia although, as we have already seen, it might actually be more appropriate to see gender as being made up of at least five sexes lying along a continuum from female to male (Fausto-Sterling, 1993).

'Gender' can be said to refer to the socially defined categories of 'masculine' and 'feminine' and according to Ahlquist (1994) there are many definitions to a person's gender; (i) genotypic i.e. chromosomal pattern, (ii) gonadal i.e. the presence of testis or ovary, (iii) phenotypic including the body habitus and genital anatomy, (iv) hormonal i.e. oestrogen or androgen effects on the target tissue, (v) legal i.e. the gender that appears on a birth certificate or passport and (vi) social which includes how an individual sees themselves, how they approach every day matters such as which changing room or toilet to use and their choice of sexual relationships.

'Gender role' is used to refer to the behaviours, interests and tasks which are socially defined as being appropriate for males or females (Turner, 1995). 'Gender identity' refers to an individual's self-concept of his or her sex - classified in our culture as 'woman'/'girl' or 'man'/'boy'

At one time it was assumed that gender was 'inborn' and rigid, however, almost fifty years ago, John Money (a psychologist at the John Hopkins University in America and a pioneer in work with individuals with intersex conditions) proposed that nurture could take precedence over nature in deciding gender (Money *et al.* 1957).

Money's work was well received and for several decades it was accepted that as long as gender reassignment took place within the period of gender plasticity (which Money felt lasted until the age of 2 years), that a child could develop without difficulty in a gender incongruent with their biological sex. (Greenberg and Chase, 2000).

In more recent years, however, a biologist, Milton Diamond challenged Money's long-standing theories and practice. He, along with others working in the field of intersexuality provided evidence that gender is not necessarily as malleable as had been thought (Diamond and Sigmundson, 1997). Others have echoed this finding that patients who have had their gender reassigned due to *physical* abnormalities (i.e. in conditions such as cloacal exstrophy where the bowel, bladder and other organs are exposed to the outside) may actually have significant problems and as many as a third may opt for reassignment back to their biological sex in later life (Reiner *et al.* 1999).

Ahlquist (1994) believes that it is the discordance between our social definition of gender (see earlier) and other definitions that can lead to problems of sexual identity in those with intersex conditions. Slijper *et al.* (2000) note that it is those girls whose gender was reassigned later in childhood, compared with those whose female sex was immediately assigned at birth who are more likely to experience a disorder of gender identity. They claim that this is due to the environment within which they are raised. It is the sex parents feel their child has which influences how they deal with the child; external appearance is one of the most important contributors to a child's gender role

identification and gender identity and this is influenced by confusion within families about the ‘real’ gender of the child (Slijper *et al.*, 2000).

When there is little or no ambiguity about the physical appearance, such as in CAIS, these children are generally raised as girls and their gender role behaviour and gender identity are said to be female (Slijper *et al.*, 1998). For children with PAIS, however, the situation is more complicated because these patients will have been born with some ambiguity to their genitalia and the parents will, therefore, have been more aware of their child as having been ‘male’ to some extent. Children with PAIS raised as girls are said to be at greater risk of developing cross-gender role behaviour and cross-gender identification (Gooren and Cohen-Kettenis, 1991).

It seems that sex assignment to the gender incongruent with the chromosomal sex may prove more successful in cases where children have been exposed to lower levels of male hormones in utero (Migeon and Wisniewski, 1998) and that here the distinct line between ‘male’ and ‘female’ is less well defined. Where individuals have conditions such as AIS, studies have shown that almost all those assigned to a gender incongruent with the chromosomal sex were content with the gender in which they have been raised. Very few showed a gender identity or gender role in accordance with their chromosomal sex (Wisniewski *et al.* 2000).

Children with a condition called congenital adrenal hyperplasia have been exposed to androgens in the womb due to a genetic abnormality, and girls with this condition, although born with XX chromosomes and female internal reproductive organs, will

show a varying degree of virilisation of their genitals at birth. In addition, there is evidence to suggest that these girls demonstrate increased masculine gender role behaviours, showing preferences for male-typical activities, toys and dress. However, girls with congenital adrenal hyperplasia tend to have a female gender identity and do not want to be male (Hines and Kaufman, 1994). It has been argued that whilst the androgen exposure may have shaped the gender role of these children, it has not been sufficient to create a male gender identity and environmental influences (i.e. being raised unambiguously as a girl) have helped to shape this female gender identity (Berenbaum and Hines, 1992).

The biological argument may go some way to explain why the children with PAIS and raised as girls in studies such as the Slijper *et al.*, (2000) research, were more likely than the children with CAIS to show masculine gender role behaviour and gender identification.

It is not entirely clear what proportion of the differences seen in gender role behaviour in children with intersex conditions are due to biological factors (e.g. the influence of hormones on the brain) and which are influenced by early socialisation (e.g. social stereotypes) (Meyer-Bahlburg, 1994). Pre and post-natal androgenisation of the brain is thought to be responsible in some degree for incongruent gender role behaviour (Berenbaum and Hines, 1992). However, it is unlikely to be the only cause - not all children experiencing this exposure develop such problems and children born with a greater level of virilisation (indicating higher exposure to

androgens) do not necessarily show a greater degree of boyish behaviour (Hines and Kaufman, 1994).

The debate about what is more important for gender identity development, the biological sex or the sex a child is reared in, continues (Slijper *et al.*, 1998); however, whatever the outcome of the debate, it seems to be the case that children with a physical intersex condition may constitute a high risk group as regards the development of disorders of gender identity (Slijper *et al.*, 1998).

Gender identity disorder is diagnosed in the general population when children show a strong and persistent cross-gender identification and persistent discomfort with their sex, or sense of inappropriateness in the gender role of that sex (American Pediatric Association, 1996). The most typical problems of gender development in intersex children and adolescents include: (i) marked gender atypical interests and behaviours that cause parents concern, (ii) body image problems associated with ambiguous genitalia or with the development of gender-contrary secondary sex characteristics at puberty, (iii) questions about sexual orientation, (iv) gender insecurity or doubts about correct gender assignment and (v) frank requests for a sex change (Meyer-Bahlburg, 1994).

Gender Assignment in Androgen Insensitivity Syndrome (AIS)

The previous section considered how views on the issue of gender identity have developed over the last 50 years or so and, with this in mind, it is important to consider how children with AIS are currently treated with respect to gender.

Children with complete androgen insensitivity syndrome (CAIS) are generally raised as girls (Slijper *et al.*, 2000) although in the partial form of the syndrome (PAIS) the gender assignment may be less straightforward. Sex assignment in this group of male pseudohermaphrodites born with ambiguous genitalia (i.e. PAIS) is a more difficult matter and that there are still no reliable criteria for their sex assignment (Slijper *et al.*, 1998).

Viner (1997) states that there is a lack of knowledge regarding the criteria used to decide the management of infants with partial AIS with respect to the sex of rearing, in fact he claims that the diagnosis and management of PAIS is surrounded by inaccuracy and confusion for parents and states that there is relatively little consistent research from a psychological point of view to guide them as to how to make decisions around the gender assignment.

Advice concerning the timing of gender assignment also varies. Diamond (1996) claims that currently no allowance is made for intersexed persons to grow up in the bodies they possess so that they can eventually decide for themselves what 'normal' sexual function is. Diamond (1996) is therefore of the opinion that any sex

assignment or genital surgery should be delayed until the child can decide for themselves which means that the child would be raised neither as a boy or as a girl but as an intersex person. This view is supported by some adults with intersex conditions who have expressed their anger that decisions were made on their behalf which have shaped their whole future, before they were able to know 'who' or 'what' they were for themselves (Holmes, 1994).

Psychiatrists and other professionals who have had contact with patients who have later rejected their early gender assignments believe that it is better to do nothing (especially not surgery) and to let patients decide for themselves when they are ready (Hendricks, 1993). However, Slijper *et al.*, (1998) state that whilst children are able to say at a young age whether or not they want to have their genital organ corrected, it is beyond the capability of a child to develop an intersex identity.

Slijper *et al.*, (1998) feel that a period of uncertainty as to gender assignment can be traumatic for the child and the parents and that children with an intersex condition should be assigned either male or female and raised unambiguously in the assigned sex. They consider that early sex assignment and correction of ambiguous genitalia are essential and later, from this safe psychological and social position and judging by their gender feelings, the children can decide whether the assigned sex is right or wrong. This means that parents would need to decide about the gender of their child at birth, however the authors feel that postponement is not a reasonable option and that a reconsideration of the sex assignment is inevitable.

Another reason that is given for early sex assignment to the most appropriate gender is to reduce the difficulty parents will have in dealing with their child's diagnosis (Slijper *et al.*, 1998). Other authors agree that gender assignment must be done as early as possible to avoid subtle rejection of the child by the family, particularly if the child's appearance is grossly discordant with the chromosomal or expected sex (Donahue, 1987). Parental feelings of shame and guilt can deprive the child of an opportunity to deal with the diagnosis (Slijper *et al.*, 1998).

In Slijper *et al.*'s (1998) study it was reported that where the diagnosis was of male pseudohermaphroditism with an unambiguously female appearance (i.e. complete androgen insensitivity syndrome), 79% of parents were able to deal with the assignment to female gender despite the incongruent chromosome pattern. However, those parents of children with male pseudohermaphroditism and ambiguous genitalia (i.e. partial androgen insensitivity syndrome) had much more difficulty coping with the gender assignment of their child. 50% of parents with such children were said to be unable to deal with the lack of gender clarity and two mothers and one father (6% of the total group of 27 couples) openly rejected their child as a result (Slijper *et al.*, 1998).

Given that individuals with AIS are biologically male but phenotypically female, this has given rise to questions about the likely sexual orientation of such individuals. Some researchers have cited increased rates of bisexuality and homosexuality in women with conditions where there has been prenatal exposure of the brain to androgens (Dittman *et al.*, 1992).

There have been several studies which indicate that women with complete AIS (and therefore an inability to respond to prenatal androgens) are no more likely to be homosexual than women in the general population (e.g. Masica *et al.*, 1971; Money *et al.*, 1984; Lewis and Money, 1986). There appears to be little literature that describes the sexual orientation of women with partial androgen insensitivity syndrome, although it might be expected that this group would show a greater incidence of homosexuality due to the degree of sensitivity to the androgens that their condition allows.

The few studies that there have been concerning the sexual orientation of women with intersex conditions, do not permit a clear assessment of the role played by either prenatal steroid hormones or postnatal socialisation factors in the ultimate expression of sexual orientation and behaviour (Pardridge *et al.*, 1982).

Outcomes for Children with Intersex Conditions

There have been relatively few detailed studies of the psychological outcomes for adults with intersex conditions (e.g. Wisniewski *et al.*, 2000) and almost none that look at the impact, if any, within childhood. Without replicated studies that provide reliable measures of the types of problems that such children experience or the factors that influence these, it is difficult to know what effect, the intersex condition has on a child's well-being.

Half (50%) of the children with CAIS in Slijper *et al.*'s (1998) study exhibited general psychopathology (selective mutism, anxiety, sexual problems, oppositional defiant disorder, depression), as did 46% of the children with PAIS. There is no indication then that general psychopathology is related to androgen effects on the brain and we must therefore consider alternative causes for the onset of psychopathology in children with intersex conditions where the chromosomal and anatomical sex are discordant (Slijper *et al.*, 1998).

The children described above had been re-assigned female within the first 4 weeks of life and, where necessary, underwent a correction of their genitalia at a young age. It seems that the early timing of sex assignment and genital surgery, whilst thought to be of key importance (Diamond and Sigmundson, 1997), is not sufficient on its own to prevent psychological difficulties for children with intersex conditions.

Such psychological problems are twice as prevalent among children who do not begin to receive psychological support and help as soon as the diagnosis was made compared to those who do (Slijper *et al.*, 1998). Early counselling seems to have a preventative effect although even adding this element to the package of care families receive does not appear to be sufficient to keep all children from developing signs of psychopathology.

The Disclosure of Conditions to Children

Parents report finding it very difficult to inform their child about their diagnosis of AIS and to offer adequate support around this disclosure (Slijper *et al.*, 2000). There is relatively little consistent psychological research to help parents with the disclosure of the details of the condition to the child themselves (Viner, 1997).

Due to the paucity of literature concerning the difficulties for parents in making the disclosure of an intersex condition to their child, this section draws on research looking at how parents talk to children about other conditions - primarily HIV and cancer. Although intersex conditions may not have the same physical outcomes for the children, there are issues of secrecy, taboo and stigmatism that appear to be common to all of these conditions and there are also likely to be parallels in the experiences of the parents.

Disclosure of HIV

Around a fifth (23%) of parents are said to choose complete non-disclosure to their children with HIV. A fifth (20%) decide to use deception (i.e. to tell their child they have a different illness), two fifths (40%) choose partial disclosure and less than one fifth (17%) choose to make a full disclosure to their child (Funck-Brentano *et al.*, 1997). However, The American Academy of Pediatrics (1999) considers that persistent nondisclosure may expose children to detrimental anxieties, leading to

emotional distress, inhibition, immaturity or maladjustment and this body strongly encourages disclosure to school-age children.

Parents' primary concern is said to be to protect their child from depressive or fearful reactions (Funck-Brentano *et al.*, 1997). However, it is reported that most children report stressful experiences related to their disorder, regardless of the degree of disclosure and that rather than causing children more distress, the disclosure of the HIV diagnosis actually helps the children to cope with their illness (Hardy *et al.*, 1994).

In addition to worrying about the impact that the HIV diagnosis might have on the child, parents may also worry about the effect that disclosure may have on their relationship and may fear anger from the child (American Academy of Pediatrics, 1999). There may also be a fear of inadvertent disclosure by the child to others outside the family group and a concern that this would lead to stigmatisation, discrimination and ostracism towards the child and other family members (Funck-Brentano *et al.*, 1997; Penny, 1999).

Penny (1999) lists a number of reasons why children who are infected with HIV need to be told: (i) so that they can make sense of any changes happening to them, (ii) so that they understand why they go to hospital, (iii) so that they feel more in control because they can ask what might happen next, (iv) so that they can ask for help or support if things are difficult and (v) so they feel more able to tell their parents what is worrying them.

Hardy *et al.* (1994) found that parents expressed confusion and uncertainty as to the most appropriate age at which to tell their child about their diagnosis. There seems to be a fairly standard pattern of disclosure to children in early infancy (below 5 years) who are told almost nothing and to adolescents (14 years and above), most of whom are told a most of the information about their condition. However, in primary school-age, children (between 6-11 years) the patterns of disclosure are more diverse (Funck-Brentano *et al.*, 1997) and there appears to be no clear age at which disclosure occurs.

The American Academy of Pediatrics (1999) recommends that HIV infection should be disclosed in an individualised manner, taking into account the child's age, psychosocial maturity, the complexity of the family dynamics and the clinical context. They state that, in general, younger children do not need to be informed of their diagnosis although they strongly recommend disclosure of HIV status to school-age children and adolescents.

Disclosure of Cancer

Similarly, in the cancer literature, most paediatricians are said to disapprove of 'deceptive attitudes' in the parents because irrational explanations and lies on their part will result in an untrusting atmosphere and may generate negative outcomes in the children (Slavin *et al.*, 1982). Other studies have shown evidence of potential adverse effects on children such as increased anxiety and isolation when they are

aware of their cancer diagnosis without being told but do not talk about it due to mutual pretence (Spinetta, 1980).

Parents may be so emotionally distraught that they are unable to talk with their child and in this case a lack of disclosure may communicate the unintended message that the condition is a morbid, frightening secret to be worried about and that it is so toxic and dangerous that it cannot be discussed openly (Claffin and Barbarin, 1991). Other studies have also emphasised how accurately children are able to perceive cues of parental distress and they may therefore realise the seriousness of their condition by parental reaction even if little or no information is communicated directly to them (Chesler and Barbarin, 1987).

It seems that nondisclosure fails to mask the salient and distressing aspects of the disorder and even though younger children are often told much less than older children about their disorder, they experience similar levels of distress (Claffin and Barbarin, 1991). Children are said to be able to sense parental distress and may realise that there is something wrong or a secret concerning them through their parents' reaction, regardless of whether the information is directly communicated to them (Chesler and Barbarin, 1987; Claffin and Barbarin, 1991).

The 'stage by stage' approach has also been recognised as the best option in the diagnosis of cancer (Spinetta, 1980) with disclosure geared to a child's level of cognitive development and psychosocial maturity (Woodard and Pamies, 1992.)

Disclosure of AIS

Goodall (1991) tells us that for a grown woman to find out that her chromosomes are male when her lifelong image has been totally female can create a painful crisis of identity likely to cause severe emotional difficulties. She argues that, if the information is available, it is much better to disclose it to protect the child from a shocking revelation at an age when conformity with peers and a stable sexual identity are most important. Indeed, some assert that a policy of secrecy will cause much more damage than the condition itself (Brooks, 1999).

Despite the general consensus of opinion today that children with intersex conditions should be told about their condition, many parents still choose not to disclose the full information to their child as recommended. Reasons for this may vary but a number of the possible factors involved may be considered.

It is likely that parents will want to prevent emotional pain to their child and they may believe that since the genetic information cannot be used in any way to correct the condition, that it is better not to tell their children about the intersex condition. They might consider that the knowledge would reduce the child's freedom to 'be themselves' (Minogue *et al.*, 1988).

In Slijper *et al.*'s (2000) study parents of children with AIS experienced an authoritative medical attitude which may have precluded them from receiving or asking for full explanations from the physicians who were of the opinion that the

parents and their children would be unable to cope with the knowledge of a 'male karyotype'. The result is that these children were badly informed and confused for years and that even after hearing the whole story about their condition some of them have found it impossible to incorporate the correct information into their distorted self-image.

It seems that there is an increased risk of developing a gender identity disorder for children with an intersex condition, and a part of the concerns parents have about telling their child of the disorder may be the fear that the knowledge will cause their child to experience a gender identity crisis (Ellas and Annas, 1988).

However, although parents may fear that disclosure will be distressing, there is evidence that non-disclosure can be more stressful for children than actually knowing the details of their condition and a policy of total non-disclosure in a family until the child reaches early teens can actually be instrumental in causing a severe emotional crisis of identity in adolescence. Knowing more can actually help a child to cope with the experience (Goodall, 1991).

A policy of non or limited disclosure doesn't serve to spare children from the salient and distressing aspects of a chronic or long-term condition and that the very experience of living with such conditions, their treatment and management and the reactions of those around them, unavoidably presents the child with information about the seriousness of the situation. The benefits of a child being told what is happening appears to outweigh any upset that may occur (Minogue *et al.*, 1988.).

The Outcomes of Non-Disclosure

Anecdotal reports from adults with AIS can provide some understanding of the impact of AIS on their lives, particularly with respect to their experience of the disclosure (or lack of disclosure) of the condition. However, caution must be exercised in considering this information. One reason for this is the marked change that has occurred within our society over recent decades in terms of the degree of openness that is present concerning all matters related to sex and gender. For adults now who grew up in a culture where talking about such issues was more of a taboo, it is possible that this, and other variables, will have had an impact on their experiences. In addition, the policies adhered to by medical and other professionals may have changed over time with relation to AIS, in particular, in relation to the issue of disclosure. The issues for children growing up today may, therefore, be somewhat different, although it is likely that many of the issues raised by today's adults with AIS will still be relevant.

Reasons given for non-disclosure have included concerns about the effect that this might have on the individual with AIS and receiving the diagnosis has been said to cause feelings of shock, grief, anger and shame for the individual themselves as well as for their parents (Slijper *et al.*, 2000). Adults with AIS report that they have sometimes felt overwhelmed by unwanted thoughts about AIS and have tried to avoid thinking about it. They find the lack of fertility particularly difficult to accept but also struggle to come to terms with the discrepancy between their genotype and

phenotype and their perceived reduced chance of finding a partner (Slijper *et al.*, 2000).

However, adults with AIS who did not receive details of their diagnosis as they were growing up, report difficulties in a number of areas. They speak of reduced self confidence with some suggesting that living in fear of a terrible secret as a child has the effect of reducing their enjoyment in every part of life and leads to reduced ability to cope with life's challenges (Kemp, 1996). Additionally, adults with AIS speak of the loneliness they have experienced, saying that to have one's diagnosis kept a secret increases feelings of 'freakishness' and reinforces a sense of isolation. To discover the truth of the disorder alone and from textbooks is said to be devastating (Anonymous^a, 1994).

Some adults with AIS speak of fearfulness and confusion, and state that whilst young girls might be happier not knowing about their male genetic or gonadal status, it is not possible to guarantee ignorance by nondisclosure. It is said to be essential that parents have the opportunity to talk openly with a professional counsellor at the time of diagnosis so that they don't become imprisoned by their own feelings which can further dissuade them from making any disclosure to their child. Non disclosure leads to children feeling isolated and confused about their gender, particularly if they are aware that something wrong but are unable to discuss it with anyone (Anonymous^b, 1994).

Shame is an issue which adults with AIS raise within the literature, warning that when parents are unable to talk about the diagnosis with their child it becomes a secret too terrible to ever mention within or outside of the family. Whilst it is acknowledged that parents who forbid disclosure may do so to protect themselves, some believe that but nondisclosure could be far more damaging for the child than knowing the truth within a supportive and caring environment (Anonymous^b, 1994).

In Slijper *et al.*'s (2000) study, a feeling of shame about their intersex condition was frequently reported by adult patients, more so than by their parents. These authors hypothesise that taboos around discussing sex, menstruation or infertility when these women were growing up are partly responsible. Additionally, the timing of the diagnosis may have been important to these older women did not receive their diagnosis even in adolescence when it became apparent that there was an absence of menarche. Adolescents with primary amenorrhea worry about the functioning of their bodies and feel different from female peers, which can cause feelings of shame. The confirmation of these worries with a diagnosis of AIS serves to enhance these feelings.

In summary then, when parents consider disclosure to their child about medical conditions including HIV, cancer and AIS, the decision about whether to discuss the diagnosis with their child is likely to be dependent on a number of factors including; the parents' own understanding of the condition, their concerns about the effect that knowing will have on their child's life, their beliefs about whether knowledge will be

helpful or damaging to the child and their trust in the opinions and advice given by those professionals involved.

When, How and What to Tell

When to Tell

In terms of child cognitive development, the literature suggests that at the age of 2 years, a child's approach to life is 'totally egocentric' (Goodall, 1991) and that until the age of 6 years they are too involved in their own internal world to be concerned with making comparisons with others. However, by 6 years children are beginning to make deductions and can sometimes draw faulty conclusions. From 7-11 years, deductive abilities improve and the making of correct connections becomes possible. In the pre-teen and teenage years, the concept of matching and therefore 'shaping up' become important (Goodall, 1991).

Children's concepts of illness causality evolve as a function of age in a systematic and predictable sequence during the course of cognitive development (Burbach and Peterson, 1986) and cognitive coping strategies are utilised by young children and become more frequently used as children become older (Claffin and Barbarin, 1991).

A child's development stage should therefore be taken into account in disclosure. Goodall (1991) states that, whereas at one time it was customary to withhold distressing information until a child was considered old enough to understand it,

nowadays it is recognised as more helpful to tell the truth stage by stage as understanding unfolds. She argues that, in any chronic disorder of early onset including those with a chromosomal abnormality, it is kinder to reveal this to the child gradually and at appropriate stages of cognitive and physical development (Goodall, 1991).

Warne (1997) agrees with this and states that a child with AIS has the right to know everything. He recommends that the information should be given in stages, taking into account the child's level of development and ability to understand what is being said. With this in mind, he recommends that between 6 and 11 years, a time where most children have little ability to think about the long-term consequences of events, they need simple explanations for the reason why they need to see the doctor. However, girls usually develop the ability to reason and think in philosophical terms around the age of 12 and it is only at this time that a girl with AIS will be able to understand a discussion about the complex nature of her condition and the implications of this. Even then, she will be quite unfamiliar with the internal workings of her body. Warne (1997) emphasises that it is important for children with AIS to know about their condition before adolescence and the pressure from the peer group to conform and not exhibit difference. Since children learn about chromosomes in school around the age of 15-16, it is best for them to be informed about their diagnosis before this time.

Slijper *et al.* (2000) recommend that children with AIS should be gradually informed of their condition, having full knowledge of their condition *except for* the chromosomal pattern by the age of 11. By the age of 16 or 17, individuals with AIS should be fully aware of all aspects of the condition.

How to Tell

Slijper *et al.* (2000) suggest that children with AIS should be informed about their condition by their own parents in three steps: (i) early education about the biological and psychological aspects of normal sexual development so that they have a baseline from which to be able to understand the ways in which their own body differs, (ii) around the age of 11 (i.e. before hormone replacement therapy commences) all information about AIS should be given with the exception of the XY karyotype and (iii) around the age of 16 or 17 disclosure of the chromosome pattern should be given (although this may need to become earlier as children are more able to gain access to such information though the internet and so on).

When full disclosure is too difficult for parents who wish to maintain secrecy about the diagnosis, partial disclosure might be a useful step for a limited period since it allows a compromise between the child's need for information and the parents' fears of disclosure to outside agencies. For intersex conditions where there will not be a progression into illness and particularly for conditions such as AIS where there are few clinical or surgical procedures required, taking some time may allow the parents to cope with the disclosure of the condition better (Goodall, 1991).

There has been a limited amount of material produced specifically for parents of children with intersex conditions to try to help them find ways in which to give a child information about their condition. 'The Baby who was Different - A Fairy Tale' by Warne (1992) is one of the rare articles produced in 'parent-friendly' language that explains clearly and simply what the diagnosis of an intersex condition such as AIS actually means and gives a medium by which to communicate this to a young child.

What to Tell

Young children need simple explanations of conditions and clear messages about how they are caused. Primary school children need more specific details of a condition, often linking the condition to its treatment. Older children need to know more to help them make decisions about treatment and to help them to explain to others if necessary.

Goodall (1991) states that whilst familiarity with medical terms may be useful in the earliest years, a thorough understanding must come later. In mid childhood diagrams can readily be transposed by imagination to the external features of one's own body, although internal imagery can be very rudimentary (many adults have no concept of internal anatomy). By adolescence, it may be possible for a child to look back into embryological life and imagining the body's internal schema becomes possible. The development of more mature reasoning will also allow the understanding that the

removal of gonads earlier in life may require hormone replacement in some intersex conditions and that fertility will be affected.

In addition to thinking about what to tell a child, it may also be important to think about what not to say. It is recommended that parents avoid talking about secrets, which can make children feel bad or ashamed. If the information is sensitive and there is a concern that the child may indiscriminately share it with others, it may be better to call the information 'special' or 'private' (Lipson, 1994).

According to some authors, to explain the diagnosis of AIS to a patient in an 'embryologically correct' manner by telling her that she is fundamentally male but with abnormal sexual development is likely to be unhelpful. This type of explanation is felt to be unnecessary - even destructive, since it invalidates one of the most fundamental parts of a person's identity - gender. It is said that the explanation offered should begin from the understanding that the patient is female and further discussion should be built on that assumption (Anonymous^b, 1994).

Adults with AIS are able to list a number of areas that they believe will be important to consider when discussing the intersex condition with a child or young adult. Helping the individual to think about what it is that actually makes a person female will help them to work through issues of their gender identity. Additionally, the absence of menarche is often experienced as confirmation of a lack of womanhood and parents will need to be aware that this issue may need further discussion as the implication 'sinks in'. For young adults with AIS, the absence of periods can be

more important than the lack of fertility despite what parents believe will be the most devastating news. Finally, it is felt to be essential that parents are aware of the need and ensure (as best they can) that counselling is made available for their child, especially when the diagnosis is given in the teenage years (Anonymous^a, 1994).

RESEARCH AIMS AND QUESTIONS

There is very little published material concerning the experience of parents of children with intersex conditions, specifically androgen insensitivity syndrome (AIS). The literature does not appear to include any studies which consider the process parents go through in deciding if, what, when and how to tell their children about their condition.

There are studies which consider the longer-term impact on children of having an intersex condition (although these have generally been either single case studies or involve heterogeneous groups of participants e.g. Froukje *et al.*, 1998). There appears to be only one published study to date considering the impact on parents of receiving the diagnosis of an intersex condition for their child (Slijper *et al.*, 2000) and the limitations of already been discussed. Finally, there are the anecdotal accounts of affected individuals (e.g. Anonymous^b, 1994) and professionals (e.g. Goodall, 1991) giving their opinion about the need for disclosure based on their experience. Given the lack of published literature in the area of this study, material from other related fields where lifelong conditions affect children and families must be drawn upon.

Current opinion appears to be that it is better for children to know about their condition and its implications and that this information should be imparted gradually (e.g. Slijper *et al.*, 2001; Goodall, 1991). However, there is no literature that describes the experience of parents in considering this disclosure and the ways in

which they approach this task. It would, therefore, seem important to consider the experiences of such parents and to examine the specific thoughts and feelings which are involved in the decision about making a disclosure to their child about a condition where the phenotype and assigned gender do not match the genotype and internal anatomy.

It seems then that the experience of parents of children with intersex conditions has been largely overlooked within the literature. Through the use of qualitative interviews with parents, this study aims to provide a better understanding of the issues, thoughts and feelings that are most pertinent to parents of children with AIS. By providing an understanding of the experience for parents, it is hoped that professionals seeking to provide guidance to parents about when, how and what to disclose, will be better informed as to the obstacles such parents must overcome to be able to achieve this. They might also be better able to offer appropriate and helpful support to guide families through this difficult process.

Rationale for the Methodology

The parental experience of having a child with an intersex condition such as AIS and the specific issues relating to disclosure to the child, is an area which has been researched only in a limited or indirect way until now. Investigating this domain would therefore seem to require a qualitative approach which allows an understanding of the real nature of the experience for any given individual to emerge (Stern, 1980).

Henwood and Pidgeon (1994) describe three strands of qualitative enquiry; (i) reliability and validity, (ii) generativity and grounding and (iii) discursive and reflexive. The first strand is said to have empiricism as its underlying epistemology and would include methods such as data display (Miles and Huberman, 1984, 1994), content analysis (Krippendorff, 1980) and protocol analysis (Ericsson and Simon, 1980). Methodologically, these approaches rely on induction to discover valid representations (Richardson, 1996, pp32). The second strand has a contextualist epistemology and includes grounded theory (Glaser and Strauss, 1967) and ethogenics (Harré and Secord, 1972). These methodologies involve the construction of intersubjective meaning (Richardson, 1996, pp32). The third strand is based in a constructivist epistemology and relies on interpretive analysis and the deconstruction of texts. This strand includes discourse analysis (Potter and Wetherall, 1987), narrative analysis (Gergen, 1988) and interpretative phenomenological analysis (Smith, 1996).

Given the type of data and aims of the research project, the two main qualitative methods which presented themselves as most appropriate for the analysis were a contextualist approach such as grounded theory or a constructivist approach such as interpretative phenomenological analysis. These methods are both said to be 'symbolic interactionist' in nature (Blumer, 1969b) and use similar methodological techniques of data collection and analysis. In both cases, new developments in substantive and formal theorising are made through the close and detailed inspection of participants' accounts.

Grounded theory aims to provide a conceptually rich, dense and contextually grounded account of participants' experiences. An individual's social world is seen to be enacted through the interplay of gestures, symbols and systems of meaning and the theory is generated by paying attention to the participants' own accounts of social and psychological events in every day contexts. The contextual specificity of participants' descriptions and meanings are attended to and through interpretative work on the part of the researcher, the construction of participants symbolic words and social realities is permitted (Richardson, 1996, pp35).

Interpretative phenomenological analysis (IPA) is also said to be based in symbolic interactionism and whilst it follows a similar methodological process to grounded theory, it is concerned more specifically with the meanings that experiences hold for the participants (Smith, 1996). It seeks to explore an individual's personal perception or account of an event or state as opposed to attempting to produce an objective record of the event or state itself. IPA has developed from the social cognition paradigm in social psychology and focuses on participants' cognitions and the chain of connection between these and their verbal responses.

A grounded theory approach to qualitative research is undertaken in this research since, as a first investigation of the area, the study was primarily concerned with revealing intersubjective accounts in context rather than making an interpretive analysis of the socially constructed meanings that the experiences hold for participants. Grounded theory has been recommended as having considerable

potential for clinical psychologists, particularly in the practitioner disciplines such as health psychology (e.g. Charmaz, 1990; Wilde *et al.*, 1993) and, overall, seems most appropriate in exploring the concerns and worries for parents of children with an intersex condition.

The Main Research Questions:

- What is the experience of parents following the diagnosis of AIS for their child?
- What issues affect parents' decisions about whether, when, how and what to disclose to their children and others about the condition?

The Broader Research Aims:

- To provide a better understanding of the issues for parents of children with AIS.
- To help professionals to better understand the parents' needs so that they can address these.
- To help in the development of protocols for (i) the way in which the diagnosis is given to parents (ii) the provision of support and information for parents (iii) helping parents through the disclosure issue.

CHAPTER 2 - METHOD

Overview

Semi-structured interviews were conducted with 20 parents (8 fathers and 12 mothers) of children with androgen insensitivity syndrome (AIS). Parents were asked about their experiences of diagnosis and treatment and the impact of AIS on their lives. There was a particular focus on the parents' thoughts and feelings about disclosure to their child and other people about the condition. A grounded theory approach was used to analysis the rich data gathered from a population which has previously been overlooked in the literature and where the sample sizes are naturally small (Eiser and Twamley, 1999).

This chapter begins with details of the recruitment process and then a description of the procedures and measures used for data collection with examples from the data. Lastly, there is a discussion about the use of the grounded theory approach and its application in this particular piece of qualitative research.

Ethical Approval

Ethical approval for this study was granted by the Great Ormond Street Hospital for Children NHS Trust and The Institute of Child Health Research Ethics Committee in September 2000 and additionally by the Middlesex Hospital Research Ethics Committee in November 2000 (see appendices).

Recruitment Procedure

Potential participants were initially identified through discussion with the consultant paediatric endocrinologists at Great Ormond Street Hospital and the Middlesex Hospital who were able to provide a list of 23 children with AIS seen in their clinics over the past 5 years. The medical records for these individuals were then requested so that the diagnosis could be confirmed and basic demographic and medical data collated.

Following this, letters were sent to the parents of these children enclosing an information sheet outlining the study and its aims (see appendices) and a letter asking if they might consider participating (see appendices). After one to two weeks, these letters were followed up with a telephone-call from the researcher to ascertain whether or not individuals would be willing to participate and interviews were arranged for all those that agreed.

Due to the nature of the condition, children with AIS may not require frequent hospital appointments and are not, therefore, in regular contact with the hospitals. For several of the potential participants, contact details were found to be out of date with address and telephone numbers having changed. This stage of recruitment therefore required additional investigation through GP and Health Authority records and from the small number of names provided, even after extensive investigation, three sets of parents could not be contacted. A further two sets of parents did not have English as a first language and an interpreter would have been required. Due to

the intensive and personal nature of the interviews, it was not felt to be appropriate (or practical) to conduct the interviews through an interpreter and thus these parents were excluded from the sample.

Because 10 of the 18 children remaining at this stage were sisters, there were potentially 13 sets of parents (26 individuals in total) to interview at this stage. of these 26 parents, one couple declined to participate in the study because they said that the issues were currently too raw for them to talk about. One father declined to be interviewed because he felt he was too busy and one father (separated from his daughter's mother) did not respond to either the letter or follow-up phone messages. Two fathers were not contacted since they had separated from the child's mother and no longer had contact with their daughters.

Participants

In an effort to fully situate the sample this section will include a description of the research participants in order to allow the reader to judge the range of persons and situations in which the research findings might be relevant (Elliott *et al.*, 1999). This will mean that some information (such as the outcomes of questionnaire measures) will be presented in this chapter rather than within the results section since it seems more appropriate, given the nature of this study, to fully describe the sample before the qualitative analysis of their data.

The Parents

The 20 participants were parents of children with AIS who had been seen at Great Ormond Street Hospital and who remained on the current patient list either at this site or at the Middlesex Hospital (where care is typically transferred when children reach the appropriate age). The intention was to interview parents individually, however, in two cases the participants requested that they be interviewed together as a couple.

of the 20 interviewees, 14 of the interviewees were from seven married couples (7 men and 7 women). of the remaining 6 interviewees (5 women and 1 man), 5 were separated from their child's other parent.

Overall, 8 fathers were interviewed and 12 mothers. The parents ranged in age from 35 years 2 months to 55 years 6 months old (mean age = 48 years and 8 months) and worked in a range of occupations. 18 of the parents described their ethnic group as 'White British' with the remaining two describing themselves as 'Pakistani'.

Twelve of the parents had only one child with AIS and the remaining eight participants had two children with the condition.

All parents completed a copy of the General Health Questionnaire (Goldberg, 1978) and only one received a score that could be considered to be within the clinical range. This indicates that the majority of the parents were not experiencing clinically significant symptoms of symptoms of anxiety, insomnia, severe depression or social

dysfunction at the time of the interview. The one mother who obtained a significant score, described great difficulties in coming to terms with her children's diagnosis and found the interview material very difficult to discuss. This parent is currently receiving support from a clinical psychologist.

(ii) The Children

In total, 17 children were represented in this study. Ten of the children represented 5 sets of sisters. At the time of the interviews the children ranged in age from 3 years 4 months to 21 years 9 months (mean age = 11 years 7 months). Fifteen of the children were described by their parents as belonging to the ethnic group of 'White British' with the remaining two described as being 'Pakistani'.

The majority of the children (14) had the complete form of androgen insensitivity syndrome (CAIS) and the remaining three had partial AIS (PAIS) and had been born with what appeared to be an enlarged clitoris. All the children had XY chromosomes and had been raised as female from birth.

Fifteen of the children with AIS whose parents were interviewed were in the appropriate age range for the use of the Child Behaviour Checklist (CBCL 2-18) with their parents. of the 15 children assessed, only one received a score in the clinical range for internalising problems and none were in the clinical range for externalising problems. The one child who scored in the clinical range had experienced significant

physical problems as the result of vaginal reconstructive surgery and a number of her symptoms were thought to relate to the impact of her medical problems.

The parents of the two individuals who were outside of the age range for the CBCL, (being 19 and 21 years old) completed questionnaires and the scores on both of these were seen to be in the clinical range. This information was felt to be useful although technically not valid (see discussion chapter for further consideration of this).

Measures

The Interview

Semi-structured interviews have been extensively used in qualitative research since they allow a relatively standard range of topics to be addressed with different individuals and allow participants' interpretations can be identified and explored in a detailed way (Richardson, 1996).

A semi-structured interview schedule was developed based on the available literature regarding intersex conditions and the impact on parents, anecdotal reports of individuals with AIS and their parents and the clinical experience of the psychologist and medical consultants involved with the intersex team at Great Ormond Street Hospital. The interview protocol comprised a set of questions and prompts which served as a guide although not all questions or prompts under a particular section

were asked in every interview. The main areas covered included (i) the diagnosis, (ii) treatment, (iii) support, (iv) disclosure and (v) impact of the condition.

In keeping with a grounded theory approach, the interview questions evolved over the course of the study, guided by the ongoing data analysis (Strauss and Corbin, 1998). For example, questions about involvement with a support group, difficulties in getting results of investigations, concerns about a child's sexual orientation and issues of guilt and blame regarding carrier status were included in the interview schedule as the interviews progressed. A complete copy of the final interview protocol can be seen in the appendices.

Questionnaire Measures

A number of questionnaires were administered as a means of describing the sample and the results of these have therefore been summarised in the previous section relating to the participant characteristics rather than in the results chapter.

Demographic Questionnaire

A brief questionnaire was constructed to collect demographic data from each participant. A copy of this can be seen in the appendices.

General Health Questionnaire (GHQ-28, Goldberg, 1978)

Each of the parents completed a GHQ-28 questionnaire relating to their own health over the past few weeks. The GHQ was designed as a means of screening for the psychological components of ill health among adult respondents in community settings (Goldberg, 1978). It contains four sub-scales which assess for (i) symptoms of anxiety/insomnia, (ii) severe depression, (iii) social dysfunction and (iv) somatic symptoms.

Although all questions in the 60 item version of the GHQ have discriminatory power, a shorter questionnaire is generally more acceptable to respondents whilst retaining good levels of reliability and validity. The GHQ-28 was therefore used in the current study with the recommended threshold score of 4/5 being used for case identification. The GHQ bimodal response scale was used to score the columns of the questionnaire (i.e. 0-0-1-1) to eliminate errors due to 'end users' and 'middle users'.

Child Behaviour Checklist (CBCL-Achenbach and Edelbrock, 1983)

Each of the parents completed a Child Behaviour Checklist relating to their child's behaviour over the past two months. The checklist is designed to be completed by parents of children between 2 and 18 years old and distinguishes between 'typical' children and those having significant behavioural disturbance (Achenbach and Edelbrock, 1983). It isolates two main syndromes: internalising and externalising but also provides specific scores for a variety of areas including aggressive behaviour,

social problems and somatic complaints. The CBCL has been well-standardised and has adequate validity and reliability (Achenbach and Edelbrock, 1991).

Procedure

When participants agreed to take part in the research an interview date, time and venue were arranged. The majority of interviews took place in the interviewee's home although five of the participants were able to attend either the hospital or university. Prior to the interview, the purpose and plan for the interview were discussed and permission to make a tape recording of the interview was sought. Participants were requested to sign a consent form (see appendices) and time was taken to answer any questions and concerns that arose.

The participant was then asked to complete 2 short questionnaires (see measures section) the General Health Questionnaire - 28 (Goldberg, 1978) and the Child Behaviour Checklist (Achenbach and Edelbrock, 1983). In addition, they were asked to complete a short demographic questionnaire. The interviews themselves lasted between 45 and 90 minutes. At the end of the interview, participants were given an opportunity to ask any further questions they might have and were informed of the expected timescale within which they would receive a summary of the research findings.

Data Analysis

Data Preparation and Storage

All identifying features (names, place names, etc.) were removed from transcripts and tapes were erased at the conclusion of the study, again for purposes of confidentiality. The data protection team at Great Ormond Street Hospital were contacted to ensure that correct procedures were understood and adhered to.

Analysis

The qualitative data analysis technique known as grounded theory was developed by Glaser and Strauss (1967) and has been refined over the decades. In the current study, therefore, the methodology follows the course recommended by Strauss and Corbin (1998).

The following section describes the process of analysis with examples from the data to illustrate each stage of the analysis. However, whilst the grounded theory methodology is described and illustrated in this chapter in a 'stage' model, this has been employed for ease of illustration. In fact, the analysis in grounded theory is more 'free-flowing' (Strauss and Corbin, 1998) and moves back and forth between the analytic techniques. In the current study, a cyclical process occurred within the analysis to ensure that the overall theory presented in the results involves a full integration of the data and is grounded in the experience of the participants.

In grounded theory, the analytic process is broken down into a series of activities to enable the data to be coded in increasingly specific ways. These will be described below and illustrated with examples from the data.

Microanalysis

In the process of microanalysis a detailed line-by-line (and word-by-word) analysis of the data is achieved by a minute focus on the text of the interview. The aim is to make the researcher aware of how much is packed into a very small portion of the data and although this type of analysis was not employed for every interview, it was used extensively at different stages of the analysis in an attempt to prevent the researcher from taking a particular stance towards the data, perhaps due to personal assumptions and bias. The process of microanalysis forces the researcher to take such assumptions into account but to consider alternatives suggested by the text and to see variations and patterns that emerge from the data.

An example of the earliest stage of microanalysis can be seen below. In the following extract the participant was describing her feelings about telling her own parents about her child's diagnosis. The extract contains only 165 words out of the 13 215 comprising this particular interview (0.01%) and illustrates just how much information and understanding can be gleaned at this early stage.

Interviewer: and what were your worries about telling them [interviewee's parents]?

Interviewee⁴: I didn't want [implies she had a choice - 'I didn't want' - a personal feeling - perhaps a decision made on her own] them [they might both worry] to worry ['worry' - perhaps quite benign - implying a sense of uneasiness. Maybe for her the term implies more serious and ongoing concern?] before they came out [idea that she might want them to worry at a later date? Perhaps after they arrive?]. and then when they came out [following on from previous statement 'and then' implies that her ideas or plans changed at this point] when she was six months [child's age - perhaps emphasising how young she was?], I didn't want [again a personal preference - in her control] to spoil their two week holiday, [belief that the disclosure of the child's condition would 'spoil' her parents' holiday] for a start, [i.e. this is only one reason for not disclosing] and also I think ... [she 'thinks' i.e. all the reason's are her opinion] I just loved her so much [strong statement of love - emphasising importance of her child to her] and she was [perhaps no longer?] my little girl, [possession - also 'little' emphasises fragility/innocence of child?] god [emphasis to support following phrase] she was only six months! [repeat of earlier expression but use of 'only' and '!' emphasise child's youth further - perhaps to justify her decision further] and I [again - personal pronoun - it's her feeling/opinion/wish] didn't want [again - she has a choice. By not disclosing she can fulfil her wants] them to be looking at her [her parents would 'see' her child in a certain way] as anything ... through anything but the same eyes that I did. [mother sees potential for others to see her child in a different way to her. Wants them to have the same view of her child as she does. Remember, she 'loved her so much' - potential that others won't love her as much because of the condition]

At the microanalysis stage, early ideas are beginning to form about the ways in which the data may relate to one another and may begin to be organised into early concepts or categories. Emerging concepts are explored in terms of their dimensions and properties, for instance, at this stage in the analysis it was felt that a category about not wanting to tell (or non-disclosure) is emerging. This category was seen to vary (at this time) in terms of the strength of the desire to withhold the information and also the reasons for wanting to keep the diagnosis to one's self.

Open Coding

The open coding stage of the analysis occurs through the conceptualisation of labelled phenomena from the interviews collected and transcribed at this point. Each transcribed interview was therefore scrutinised with notes being made in the left hand margin of the themes, issues and problems raised by the interviewees. Possible recurrences of and connections between these early themes were noted (in the form of researcher memos in the right hand margin), but were not yet systematically worked out or formalised. The aim at this stage was to remain focused on the data without making interpretations based on the researcher’s assumptions or bias.

<i>Not wanting to worry others with the diagnosis</i>	Interviewer: and what were your worries about telling them?	
<i>expression of love for child - gorgeous</i>	Interviewee⁴: I didn’t want them to worry before they came out. and then when they came out when she was six months, I didn’t want to spoil their two week holiday, for a start, and also I think ... I just loved her so much and she was my little girl, god she was only six months! and I didn’t want them to be looking at her as anything ... through anything but the same eyes that I did. I didn’t want them ... I wanted her to look gorgeous and for them to fall in love with her and not to think of her as ... maybe being a boy, or anything.	<i>Different reasons for non-disclosure: (i) not wanting to worry others (ii) not wanting others to view child differently</i>
<i>fear that others would see child differently to her if she tells them</i>		
<i>people might see the child as a boy</i>		

This process was repeated for each interview and groups of quotes from different participants were gathered together into member categories (Pidgeon and Henwood, 1996) which described a particular phenomena for this group of participants.

Whenever an extract was found which did not fit into a current member category, a new member category was created. In the analysis of data for the current study, some 39 member categories were identified. The following gives an example of just one of these:

Member	REASONS NOT TO TELL
Category	
Subheading	<ul style="list-style-type: none">• They Will Worry
Extracts	<p><i>'I didn't want them to worry before they came out. and then when they came out when she was six months, I didn't want to spoil their two week holiday ...' (P4)</i></p> <p><i>I really thought 'Why bother her?' it's not her problem. I adore my mother and I adore my father ... they're really super parents. You don't want to burden them with something. (P13)</i></p>
Subheading	<ul style="list-style-type: none">• They Will See the Child as Different
Extracts	<p><i>I just think that they're ... they're entitled to privacy. I mean they are going to ... umm ... stand out. It's different, it's something that's really different and it's something people will talk about. (P2)</i></p> <p><i>and I didn't want them to be looking at her as anything ... through anything but the same eyes that I did.' (P4)</i></p>
Subheading	<ul style="list-style-type: none">• It's her Business
Extracts	<p><i>and very early on we decided that ... we talked about the aspects of sharing this with other people and decided, for the girls' benefit, we wouldn't put word around ... about it and then they could decide in older age whether they wanted their friends to know. (P12)</i></p> <p><i>If she decides when she grows up and she's got a mind of her own and everything, that she wants people to know, then it's down to her and she can tell people ... but we didn't feel it's our decision to tell people about what condition she's got, you know ... it wouldn't be fair on her. (P19)</i></p>

Axial Coding

The axial coding stage takes place once theoretical saturation has been achieved (i.e. no new member categories or themes emerge from the current data) and allows the researcher to concentrate on devising researcher categories (formalised groups of member categories which appear to exhibit common themes) and considering the relationship between such researcher categories.

Initially, in the current study, one researcher category was developed to encompass the issues surrounding the decision to disclose. However it became clear that one category would not be sufficient to describe this phenomenon since there were some quite different issues driving the decision making process for parents depending on whether it related to their child or to other people. For this reason, the category was separated into two researcher categories; ‘deciding whether to tell the child’ and ‘deciding whether to tell others’.

Once researcher categories and member categories have been identified, the task is to group similar researcher categories under a common heading or *classification* (Strauss and Corbin, 1998). Data which may previously have been *fractured* during the open coding stage may now be reassembled so that more precise and complete explanations of the phenomena may be given. For example, the subheadings seen in the example below were gathered together to describe the two member categories, which in turn define the researcher category.

Researcher Category	(3)	DECIDING WHETHER TO TELL OTHERS
Member Category	(i)	REASONS NOT TO TELL
Subheadings		They will worry
		They will see the child as different
		People will be cruel to the child
		It's her business
Member Category	(ii)	REASONS TO TELL
Subheadings		They were with us at the time of diagnosis
		Need close family to know for support
		Regret telling some people now
		Find people who can be trusted
		Some people need to know

Overall, 39 member categories and 11 researcher categories were identified and at this axial coding stage of analysis, it became clear that a third of the categories defined in the open coding stage referred specifically to the idea of ‘disclosure’ (i.e. telling the child and others about the condition). Since this seemed to be such a significant element of parental experience it was decided that it would be better to analyse the data in two parts - one focusing purely on the issue of disclosure and the other relating to the overall parental experience from the time of receiving a diagnosis of AIS onwards. Thus, the analysis in this study involved two grounded theory accounts of the data.

Selective Coding

Selective coding is the final stage of the analysis and occurs when all researcher categories are considered to be saturated (i.e. no new member categories emerge during coding to define the researcher categories further). It is only at this time that the major researcher categories can finally be integrated to form a larger theoretical scheme or, in other words, the research findings take the form of theory. Once the data were reduced from many cases into concepts (or categories) and then classified, these were integrated and used to explain the experience of the parents in a more general, theoretical sense.

Deciding on a core or central category involves condensing the products of all the analysis into a few words to give a logical interpretation of what the research is 'all about' (Strauss and Corbin, 1998). Tools used at this stage included reviewing and sorting through memos and writing a storyline (Strauss and Corbin, 1998) to give a general gist of the research procedure and outcomes. From this, it is then easier to stand back from the details of the research and deduce the main, overriding themes. In addition, consultation with colleagues and both academic and clinical professionals gave the possibility of 'brainstorming' exercises to generate ideas as to the central category of the research.

Credibility Checks

As recommended by authors such as Elliott *et al.* (1999) and Greenhalgh and Taylor (1999), several methods were employed in the current study to verify the credibility of the categories, themes and accounts that emerged.

Two other experienced clinical psychologists acted as ‘analytical auditors’ (Elliott *et al.*, 1999, p.222) and reviewed the data for discrepancies, overstatements and errors. The first of these was a principal clinical psychologist working as a paediatric liaison psychologist for the endocrinology and urology specialities at Great Ormond Street Hospital and regularly seeing children with intersex conditions and their parents. The second researcher was a clinical psychologist and a senior lecturer in the Sub-Department of Clinical Health Psychology at University College London (UCL) with a special interest in qualitative research and experience of grounded theory methodology.

At each step of the study development, from the creation of the interview schedule through the data collection process and at each stage of the data analysis, the procedure was checked and suggestions for amendment and improvement incorporated. The categories that emerged were thus verified in terms of their methodological consistency and their clinical validity.

When the grounded theory analysis was complete and the categories established, the original researcher took a further ‘verification step’ (Elliott *et al.*, 1999, p.222) of reviewing the data in detail to check for discrepancies, overstatements or errors in the categories as they had been defined. At this stage, several categories were adjusted slightly to clarify the content and to ensure all views had been accurately represented.

Owning One’s Perspective

There is no way of completely abolishing or fully controlling for observer bias in qualitative research (Greenhalgh and Taylor, 1997). All researchers should demonstrate ‘reflexivity’ or sensitivity to the ways in which the researcher and the research process have shaped the collected data (Mays and Pope, 2000). It is said that by providing a description of their theoretical, methodological or personal orientations, researchers allow others to interpret the results of the study accordingly (Elliott *et al.*, 1999).

In terms of clinical perspective, the researcher in the current study had a particular interest in child psychology and hoped to work in this field on qualifying. During training, one year was spent in a specialist children’s hospital involving work within the paediatric liaison psychology department linked to the endocrinology and urology specialties. As part of its remit, this team provided care to children with intersex conditions and their families. The researcher did not undertake any direct clinical work with children with AIS and their families, but did attend regular intersex clinics with the medical teams from both specialities and psycho-social ward rounds with

the nursing team on the main urology ward. The current study grew out of the experience of working with these teams and a growing awareness of the lack of research in the concerning the experience for parents.

At the outset of the study, the researcher was thus familiar with the clinical nature of androgen insensitivity syndrome but held relatively few preconceived ideas about the general experience for parents when they had a child diagnosed with this condition. It was expected that many would have found the experience difficult to cope with due to the 'strangeness' of the information and the incongruence of the physical appearance with the child's chromosomal sex. It was also felt that many would perhaps be reluctant to talk about their experiences, thoughts and feelings and that privacy or secrecy would be an important issue. In terms of the disclosure issue, the researcher supposed that this would have been a universally difficult experience and that the majority of parents would have told their children very little at all.

CHAPTER 3 - RESULTS

As was described in the method section, the grounded theory analysis of the study data was divided into two main sections: (i) The Experience of Parents from Diagnosis and Beyond and (ii) The Issue of Disclosure. The results of each of these sections will be presented in turn.

For both parts of the analysis, each researcher category is described through a description of its constituent member categories with extracts from the data (quotes) to support these themes. The participants' words are presented in italics and participants are identified by their research number. There appeared to be very few systematic differences between the accounts provided by mothers compared to fathers and therefore subjects are not identified by gender.

Table 1

Being a Parent (From Diagnosis and Beyond)

Central Category		POWERLESSNESS				
		<i>'I'll be honest with you - at the moment we parents are lost. That's the fact.'</i> (P17)				
Classification		(A) The Diagnosis		(B) The Medical System	(C) Implications of the Diagnosis for the Child	
Researcher categories	(1) Receiving the diagnosis - the experience for parents	(2) Needs following diagnosis	(3) Impact of and adjustment to the diagnosis	(4) Experience of the medical system	(5) Issues of Gender	(6) The future
Member categories	i. 'A Bolt From The Blue'	i. The search for information	i. Rejection	i. Good Doctors	i. Physical Appearance	i. Expecting Trauma
	ii. Memories and Misconceptions	ii. Feelings about counselling	ii. Grief About Infertility	ii. Bad Doctors	ii. Behaviour	ii. Relationships
	iii. Emotional Impact of the diagnosis	iii. Feelings about the Support Group	iii. Guilt and Blame	iii. Challenging The Doctors	iii. Gender Identity	iii. Sex Life
		iv. Talking to someone who really understands	iv. 'It Could Be Worse'	iv. Being Guinea Pigs	iv. Sexuality	iv. Restrictions on Sports
			v. Having Another Child	v. Decision Making		

SECTION 1: BEING A PARENT (FROM DIAGNOSIS AND BEYOND)

(A) Parents’ Experience of the Diagnosis

(1) RECEIVING THE DIAGNOSIS

This category looks at the moment where the diagnosis of androgen insensitivity syndrome (AIS) was given to parents. It considers their reaction to receiving the diagnosis and the experience of undergoing periods of tests with their child to confirm this. It also considers what parents understood of the condition from their often brief consultations with the doctor to receive the news and their immediate thoughts for the future following this.

(i) ‘A Bolt from the Blue’

This category describes the actual moment for parents of hearing the news that their child has AIS. It encapsulates the feelings of shock and confusion that were universally described in the interviews by parents redirecting what they perceived to be very strange information indeed.

For the majority of parents in this study, the first suggestion that anything might be wrong had been the appearance of a hernia in an otherwise healthy child and this usually occurred within the first few days, weeks or months old. However, for those parents of children with PAIS, the problem was immediately obvious due to the

ambiguity of the genitalia, and these parents talked about a period of confusion immediately after the child was born.

The baby was took and all we could see was more doctors coming in and sort of surrounding it so ... we knew something was wrong but we didn't know what was wrong. (P6)

However, no matter when the condition was first suspected, all of the parents in the study described how totally unprepared they were for the diagnosis and what a great shock it was for them to hear it.

... and he told us then and we'd never heard of it or ... and it was just, you know, a bolt out of the blue. (P3)

Given how shocking the information about AIS was for the parents to hear, it is perhaps understandable that many commented that, at the time of diagnosis, they were simply unable to process and understand the information they were given.

The trouble is it wasn't ... while we were at (hospital) it didn't really sink in. I mean the only ... no, it didn't really sink in. (P10)

Almost half (eight) of the parents interviewed had two children with AIS and thus talked about their experience of feeling that they intuitively 'knew' the diagnosis the second time around, before any tests had been carried out. Often the parents were

persuaded by a physical sign that would otherwise have caused them little concern. This category is quite the reverse of the earlier theme describing parents' complete lack of awareness of the possibility that the condition could occur.

Well that was the first thing they said in hospital in (place 1) when (younger daughter) was born. The nurse took her off to clean her up and said 'gosh she's got really soft skin!' and this nurse didn't know anything about it and we looked at each other and we knew straight away.' (P4)

(ii) Memories and Misconceptions

Parents described their recollections of what they were told at diagnosis. It is impossible to know what information the doctors actually gave at this consultation, however, the parents' understanding of what they were told often a confused or partial account of the diagnosis.

The previous categories describe the shock that parents' experience on hearing the news and their inability to take in information about the condition as a result of this. It appeared that what parents did take away were particular phrases that 'stuck' in their minds. For most parents, what they remembered most vividly (and sometimes the only thing they can recall) was being told that the gender that they had hitherto considered their child to be, did not correspond with their biological sex.

One thing which I didn't like ... didn't like ... I was very much concerned when (Doctor) told me that, you know 'They were born as boys.' (P17)

Parents described this news as being impossible to understand and talk about their confusion and disbelief at what they are being told.

I thought they had all gone mad! What were they talking about? She was so obviously a little girl' (P7)

It seems that what parents took in at the time of diagnosis, however confused, was the only information they have, even many years after. At the time of the research interviews, parents were rarely able to give an accurate explanation of the condition which included all aspects including (i) the chromosome picture, (ii) the presence of gonads or testes, (iii) the lack of female internal organs such as a uterus and ovaries and (iv) the production of but insensitivity to testosterone.

In terms of the label that parents felt they were given for the condition, it seems that most for most parents the condition was given the name of AIS. However, for others (particularly those with older children) the condition was given its older name of testicular feminisation syndrome and some parents were unclear as to the name of the condition at all.

I don't ...I don't even know the name of the condition. I knew the name when I read your letter! You know, because ... err ... there isn't any literature and, come on, when you go the doctor you hear a lot of things, you don't ... you don't remember everything. (P17)

Even at the time of interview, although most parents were aware that their child's chromosomes were unusual in some way, probably only around half of the interviewees reported having a clear awareness of their child's sex chromosomes being XY compared to the usual female picture of XX. Some parents were not even clear whether it was their own or their child's chromosomes that differed from the norm:

They did say about the chromosomes but I can't actually ... I know they said ... erm ... she had both sets. (P11)

That's the thing I find quite confusing is the chromosome side of it because apparently mine were mixed or something or one was ... erm ... one of my chromosomes was deformed or ... I can't remember. (P16)

Confusion concerning the child's internal physical state also arose. Whilst all appeared to be aware that their child had no uterus, understanding of other physical implications varied enormously. Most interviewees were aware that their child had been born with internal testes rather than ovaries, however, some remained confused about this and others were unclear about other physical implications.

Well one thing I remember is that apart from the condition itself, the ... we had no idea ... we just thought it was a visual thing ... a visual deformity. Apparently it was tubes and that that they operated on ... which we was totally unaware of. (P19)

Another aspect that many parents were unclear about was the transmission or cause of the condition. Whilst a few parents were aware of all aspects of the inheritance or spontaneous genetic mutation causing AIS, more appeared unclear about how the condition had come about or the likelihood that further children or family members could be affected.

Because I think I said to them 'How did it happen?' and they said it was a mutation on the egg whilst I was pregnant ... I mean, I've never heard of that either so it's not, you know ... I just thought it would happen every time I was pregnant. (P11)

(iii) Emotional Impact of the Diagnosis

Under this member category, parents' emotions on receiving the diagnosis are considered. It seemed that for the majority, the major emotion was of being upset - even devastated by the news.

It was really devastating because ... erm ... I don't remember the exact words he said but I remember the whole room going round, you know, I was just like ... you know when you just can't focus and I was so upset and I think I started crying ... (P17)

Other parents, however, describe a state of denial or disbelief which caused them almost to feel numb to the diagnosis.

and ... err ... frankly I ... err ... I thought they might have made some mistake. I couldn't believe it. It's so earth-shattering. (P12)

(2) NEEDS AFTER DIAGNOSIS

This category deals, chronologically, with the next period of life for the parents in the study after they had received the diagnosis and then returned home with what one participant called 'mind-blowing' information and considers how parents described their needs over the following weeks, months and years.

(i) The Search for Information

Many parents expressed a need for information, either in written form or verbally given by the medical team, and they talked about their frustration at the lack of such material available to them.

We've been to (Hospital) many times, met several consultants etc. and ... err ... no-one actually said, 'Look, come on, let's sit down and talk about this' or, you know, 'Let's tell you what this is' or 'Look, go away and read this book etc. and it will tell you all about it.' (P20)

Parents described specific issues that concerned them and which they felt could have been addressed through discussion and literature. This included wanting to be able to give their children the correct information, a desire to know about the cause of the condition and its inheritance, and a wish to have some idea of what to expect for the future; both in terms of medical treatments and long-term outcomes.

.. and following on from that period is 'What is this thing?' You know, 'What's she going to turn out to be?' and ... erm ... one doctor ... showed us photos of ... you know, the sort of typical photos where the eyes are blocked out and ... thinking 'God! Is that what she ...?' and no-one's really clear about what's ...' (P5)

Many of the parents had found information from other sources. A few had found journal articles or information from medical books and a small number had used the internet to search for information. By far the best source of information, however, appeared to be the AIS support group. Parents' views about the support group in terms of emotional support will be considered in a later category, but it is clear that in terms of information seeking, the support group was the best (and perhaps the only) source of material for most parents.

It's [the support group] given us a lot of information because they have ... erm ... sort of once ever six months they do a work-sheet of ... erm ... just information and sort of ... (P6)

(ii) Feelings about Counselling

Parents appeared divided about whether or not they would have found counselling useful in helping them to cope with their child's diagnosis and the impact that this had upon their lives. Some of the parents described a definite need to talk to someone during this period and a frustration at their inability to find anyone.

I think for the first five ... when she was first diagnosed, for the first five years whilst she was going through her operations ... erm ... I really felt that we could have done with that little bit of support. (P9)

Other parents, however, expressed an opinion that they would probably not have found this useful or wished to pursue it at this time. For some this was because they found the diagnosis too difficult to talk about in the early days and for others it was because they felt that they would not have been comfortable talking with a stranger. A third reason cited was a belief that professionals simply wouldn't be able to help.

Well there doesn't seem any point in talking to other people about it who wouldn't be able to help us anyway really. (P1)

I don't think personally ... erm ... I necessarily would have wanted it ... erm ... I mean it was extremely upsetting at the time and ... erm ... (P8)

However, whatever parents' feelings about the benefits of counselling at the time of diagnosis, many of them expressed a specific need for help when their child was older - usually in relation to the idea of disclosure. They indicated that they felt the need for someone to help them think through this difficult stage.

I think it's more later when you ... when you start worrying about disclosure. That's when you sort of seek help isn't it? and counselling. (P4)

(iii) Feelings about the Support Group

The AIS support group (AISSG) was set up in the late 1980's, initially to put families in contact with one another and then to provide peer support for individuals with AIS. It launched its news-sheet (A_LI_AS), held its first meeting in 1995 and now holds these on a twice yearly basis. They developed a website in 1998:

(<http://www.medhelp.org/www/ais/>).

Almost all of the parents in the study had heard of and expressed opinions about the usefulness of such a group. Parents found the group useful for gaining information about the condition and a few parents were complimentary about the group in general. However, the majority of parents who talked about the group found that it was either unsuitable or unhelpful for them and did not meet their needs.

Because ... erm ... they're [the meetings] not really ... we don't feel they're geared to us so much at the moment. They are more to ... older women that have only recently found out, or found out sort of fairly late in life ... haven't had a chance to ... you know, get it off their chest. (P3)

(iv) Talking to Someone Who *Really* Understands

Parents were aware that AIS is a relatively rare condition and they had certainly never heard of it before receiving the diagnosis. They felt quite isolated in their situation and often had no idea of how they might go about finding out more.

and you feel like you're the only one it has ever happened to ... because you never heard of it before. It's never been in any ... papers or ... you know, it's something you've never read about. (P6)

Only two parents in the study reported actually having met another parent with a child with this condition but many of the parents talked about how much they desired this opportunity to meet with someone else in a similar position to themselves. They felt that only other parents would really be able to understand what it was like to be in their position and to offer them the support, information or advice they needed.

So I really wanted to talk to parents who had the same problem because it's OK talking to your parents, your friends, but they don't understand. If you talk to the people who the same condition, that's different. So I really wanted that. (P17)

These feelings of isolation and the unmet need of wanting to talk to other parents appeared to prompt some interviewees to want to be able to help other parents in the future. A number of interviewees expressed their willingness to help others in a similar situation by talking with them or sharing their own experiences.

I mean I'd be quite happy to ... erm ... you know, if anybody else wanted to sort of, you know ... if any other person found it helpful ... erm ... I wouldn't mind at all.
(P8)

(3) IMPACT AND ADJUSTMENT

This category integrates a number of themes which describe the immediate and short term impact that the diagnosis of AIS had on parents. These include the immediate losses that parents experienced but also their emotional journey through this early time. It also concerns the immediate impact that the diagnosis had on parents' relationship with their child and on their decision to have further children.

(i) Rejection

A number of parents explained that, following the diagnosis, they had experienced some difficulties bonding with their child. Although in all cases this appeared to be a relatively temporary problem, nonetheless it highlights a specific area of impact that the diagnosis may have.

I think my first reaction was, you know, sort of 'Get rid of it' type thing almost. It was ... it was really that, you know, that I'd had this feeling that I'd ... the way it was presented to us was that we'd created this 'creature' ... and it was that ... I mean I was quite shocked at my reaction to it, you know ... 'I just don't want it' type thing.
(P5)

(ii) Grief about Infertility

Many parents talked about the impact that the discovery of their child's infertility had had upon them. For many, it was the single most significant factor in terms of thinking about the impact of AIS. Although some reported coming to terms with it over time and finding other ways to think about the issue (e.g. 'she can adopt'), most acknowledged that the fact that their child could not have her own children had been a devastating blow to them as a parent.

All I could think about was, you know, they can't have children. (P3)

In addition to their sadness at their child's inability to have her own children, parents talked about their personal loss - the loss of expected grandchildren.

... that ... erm ... you know, that he wouldn't have grandchildren. I'd think of Christmases and things and, you know, we won't have a little family. (P2)

(iii) Guilt and Blame

This was perhaps one of the most major categories in this section of the research project. Most parents expressed some notion of guilt or blame for their child's condition and the consequences that it had already had and might have for the future.

The category concerning the receiving of the diagnosis illustrated the fact that many parents were not clear about the causation and transmission of AIS. Feelings of guilt and blame towards their child were often the result of a poor understanding of the condition and its transmission.

I mean I did tell you how sick I was to actually carry (daughter). I actually thought because she wasn't getting nourishment, that was part of the reason why she got it. She didn't develop because I didn't get any nourishment to feed her, you know? (P9)

Most of the mothers in the study talked about the fact that AIS had probably been inherited from their side and spoke of their guilt at having passed it on to their children, even though in most cases they hadn't known that they carried the condition.

Well you do feel guilty because ... you want to ... you would give them your right arm, your left arm, your everything and you've ended up giving them AIS! (laughs)
So there is that, and, of course, I've made (daughter) a carrier. (P13)

A number of the fathers acknowledged their wife's feelings of guilt over the inheritance of the condition, however, all stated that they did not feel blame or anger towards their partner.

But it's not something I ever think about. It's only when she mentions it and I think 'Well hang on ..' I just don't ... I just don't think about that at all. (P5)

(iv) 'It Could Be Worse'

A number of parents described feeling quite overwhelmed by the diagnosis and its implications and being able to think of little else, especially in the time immediately after the diagnosis. However, in thinking about the diagnosis, parents frequently expressed the view that 'it could be worse'. This thought seemed to be helpful to parents in coping with the condition. Once they were aware of the implications of the diagnosis, they were able to compare this to other childhood diseases and conditions and actually felt that they were better off than many others.

I was in hospital with mothers who were ... their children were having their heart transplant operations and ... open heart surgery, and I'll tell you something, you meet such raw courage. It was nothing compared to what those mums had gone through. (P13)

(v) Having Another Child

Having a child with AIS had an impact on parents who had planned to have more children following this baby and the diagnosis often caused them to reassess their feelings about having another child. Fifteen of the parents in the study had gone on to have further children after the child with AIS was born and, in most of those cases, the parents had known about their current child's diagnosis before going on to conceive their next. The decision to have another child, however, was never taken lightly.

We left it for a while and then when we decided we were ready to have another one ... err ... I think we saw people here ... I can't remember ... paediatricians and people, and we realised that the only thing we could do really would be to have an amnio ... and we just thought in the end 'Well it's not worth risking it' because if we found out ... by then we'd decided that if we found out ... all you could find out really was that it was XY or XX I think, and you can't just about every XY child ... you just couldn't kill a child for having AIS. (P4)

Only a few parents in the study indicated that they would not consider having any further children despite having previously expected to extend their families.

It's held me back in a way because I always said I wanted ... you know, more children ... and I won't do it again because I didn't know why it happened and if it would happen again. (P11)

(B) The Medical System

(4) PARENTS’ EXPERIENCE OF THE MEDICAL SYSTEM

Parents’ described their experience of the medical and other professional people with whom they have become involved following the diagnosis of AIS. Whilst they did have some positive stories to tell about the professionals they had met, there appeared to be far more negative accounts in the data from parents’ first-hand experiences and impressions.

(i) Good Doctors

Parents praised those doctors and professionals whom they perceived as having a ‘good manner’. Such people were generally regarded highly by parents who felt able to relax and talk to such doctors and felt listened to in return.

I can’t think of the male doctor’s name - he retired. He didn’t talk very much but he was charming and I felt easy with him. (P12)

Parents also talked specifically about doctors who were perceived as supportive. Often this just meant that the doctor provided ‘professional’ reassurance that the condition was not so serious and that the children would be fine. For other parents it meant having their decisions respected and supported.

You just want reassurance - 'Look it's OK. It's OK if they can't have children. They can adopt, they can do whatever they want'. You want somebody to ... just to say to you again. I know all that but from a professional person you hear these things and it calms you down. (P17)

Parents appeared to greatly appreciate those doctors who had time for them - perhaps over and above what they would normally expect - and also those doctors who remembered them over time and kept in touch.

and I phoned up (Doctor) ... erm ... through the support group because we hadn't met him, and he was very good. He phoned back about half past eight one evening and we had a long chat. (P2)

Additionally, those doctors and hospitals that were knowledgeable about AIS and provided parents with the information they needed were rated highly, as were those who admitted that they did not have all the answers from a medical point of view and were prepared to refer parents on if there might be any benefit in this.

The person who was in charge at that time was (doctor) and ... erm ... I sort of compiled a great big list of questions for him which ... erm ... you know, he answered ... he answered very well and I thought, you know, he dealt ... he dealt with us very well and I thought the hospital dealt with us ... erm ... extremely well. (P8)

(ii) Bad Doctors

However, despite the positive comments made by parents during the interviews, many more of them described negative experiences with doctors. In terms of the doctors' manner, parents reported that they felt looked down upon or disliked and felt that the doctors were often quite disrespectful or unfriendly.

Afterwards I thought I should have said to him 'Can you look at me?' (laughs) but you don't feel you've got that right really ... being a patient and they're ... consultants. (P11)

A number of parents felt that the doctors' manner and behaviour might sometimes be due to their feeling inadequate in terms of their knowledge or unable to deal with parents' emotional reaction to the condition.

They suddenly find out what it is, they recognise it and they think 'Oh, I can't handle this' and because they can't handle it, they don't speak to the parents. (P13)

A major criticism was that doctors could be insensitive, either in the way they dealt with the parents or in terms of the information they discussed, often in front of a child who didn't yet know the full details of their condition.

I think I could have done without (Doctor's) comments [in front of the child] of, you know, 'Tell her the whole story' (P1)

Parents also perceived the doctors they had dealt with as being deceitful at times. This ranged from discovering that the doctors had had suspicions about the child's condition which they withheld early on, to feeling that they had been told outright untruths.

The doctor there obviously suspected but didn't say anything to us because he ... took a blood sample ... erm ... (P3)

Additionally, parents felt 'fobbed off' by doctors and were often not sure what information was still being withheld from them. A number of parents described wanting to know the whole truth but feeling that the doctors had control over how much information they could have.

I don't know quite what I want to say really but ... it's ... are you told everything that they know? Because he's admitted to us that there's things we don't know, so we can't really tell you. It's a bit hit and miss. (P18)

Parents also reported feeling 'let down' by the professionals caring for their child. One such problem that parents reported was of preparing themselves and their child for a medical examination or procedure that was then not undertaken by the hospital. Another area of frustration and disappointment for parents concerned problems with test results which were lost or never fed back to parents. This included the taking of skin or blood samples from themselves and their children for genetic testing.

They asked me, could they take a piece of skin for DNA testing? and from that they would be able to ... I'm really cross about this actually ... they would be able to tell me ... if I carry this and if it's carried through my family. Now I've asked countless times for the results of that test and now they've told me ... erm ... no they didn't ... they didn't do anything with that. If I had known that they ... had on intention of telling me anything from that, I perhaps wouldn't have put her through that (P7)

Another aspect of this category is that of 'cock-ups', something that a number a parents talked about. This included doctors giving information to children in a way that caused distress, notes that were lost and even information being sent to the wrong address.

So I just sort of said to her 'I just want the information. Could you get me any leaflets or sort of brochures or whatever? She posted them through next door, which I was not impressed about. She came around, I wasn't in, she posted it through next door. (P11)

Parents also talked about the difficulties they had experienced with professionals who hadn't heard of AIS or who had very limited knowledge. For many parents, there was no-one in their local area that they felt understood about the condition and often it seemed that their local services were not even prepared to make an effort to find out about AIS.

The thing is ... err ... one thing I find really frustrating is that ... err ... a lot of the medical profession have never heard of it. and they still haven't heard of it. and when we moved to a different area and I went to see our ... erm ... GP and ... erm ... you know, he looked very blank and I had to sort of, more or less explain. (P8)

Parents seemed to find it even more frustrating when those doctors who were said to be experts were not always able to answer parents' queries. It appeared that they felt quite hopeless when faced with the fact that there might not be anybody who had the answers to their questions.

I actually felt like they didn't know! It was sort of like a guessing game to them as well, you know. (P9)

(iii) Challenging the doctors

Parents' opinions of the ways in which doctors and other professionals have dealt with them have been described. This category contains quotes from parents who have 'stood up to' (P5) the doctors. For some parents this meant refusing to be 'fobbed off' (P18) and demanding that the doctors provide them with answers to their questions.

If you ask a straight question ... erm ... the doctor can't always be a politician and answer you back because you'd ... I'd pursue it. I'd say 'What are you saying? Tell me what you mean. Don't say ...' (P18)

For others it meant finding out about the treatment options and then requesting a referral to their preferred consultant or asking that their child be considered for such treatment.

I just ... umm ... went with a support group letter to my GP and said 'I want to go and see him!' and our doctor said 'Fine' and wrote a letter. (P2)

Some parents disagreed with the doctors' opinions and refused to allow their children to undergo certain procedures that they felt unnecessary.

So I said 'No, this is entirely up to my girls when they're ready for it' and I put my foot down. He kept disagreeing with me - you know I wasn't being rude or anything. (P17)

(iv) Guinea Pigs

Parents spoke of a growing perception that their child's condition was of interest to doctors because it was something unusual and that appointments and procedures might perhaps be organised for the benefit of the medics rather than primarily for the parents. One example of this was the experience of having many doctors present at a consultation or a whole host of people 'popping in' to see the child during a stay in hospital.

Well a lot ... I just ... even now I can remember nurses coming in for no obvious reason other than to look at the baby ... and that was actually in front of us, wasn't it? (P19)

Another aspect of this was seen in the taking of samples for testing at another centre. Parents felt frustrated by the lack of feedback about the outcome of such tests and concluded that the doctors had overlooked their needs in the desire to research the condition.

You sort of think they're doing it for you for diagnosis and all the rest of it, but then you realise later, when you haven't had the results two years later and you have to really chase them up, that it wasn't really for you, it was for them! You know, it's rather annoying! (laughs) (P4)

(v) Decision Making

The parents in this study all described times when decisions had needed to be made relating to their children's care. They talked about who took responsibility for those decisions and how much they felt they were involved in such processes. Where they had made decisions, they outlined their reasons for doing so - the triggers to action. Parents talked about their feelings, with hindsight, about decisions that had already been made and their indecision concerning current and future issues.

A number of parents talked about their perception that decisions were taken out of their hands and made by the doctors.

and you're getting told from the hospital you do have a choice, and then after sort of more and more questions we were asking them ... you didn't really have a choice. You know, it was 'Well we can do this but it ... it won't be very good and it won't, you know ...' and he did say in the end 'We can overrule, you know, your decision.' (P6)

Other parents, however, described being at a loss as to what to do and having to rely on the doctors to do what they thought was best.

So in that respect, if you've got these surgeons and specialists telling you 'This is ... we need to do this', then you really sort of go with them. You know, it's ... you have to take advice from somebody, and why not a doctor? (P19)

Parents reflected on the decisions that had been made so far in their children's care and many asserted that they had no regrets over the routes they had chosen or had been chosen for them. Others expressed uncertainty at whether the decisions made had been for the best.

If their testes are in until adulthood, then they do release some oestrogen equivalent so ... erm ... that actually would be an advantage. I mean, looking back we might not have got rid of their testicles as quickly. (P2)

Where parents had made a decision, (usually about the removal of the gonads) they talked about their reasons for choosing this option. Often, the reason given was that it was better for the child to have surgery when young.

My attitude was I want it done whilst she's young so she didn't have any psychological things (P13)

Another reason given was the fear that the gonads would become cancerous if they were not removed.

Oh yes, it was discussed in detail and it was indicated that if they weren't removed, then they might become malignant so ... err ... yeah, there didn't seem an option there. (P8)

Some parents also acknowledged that a primary reason for wishing their child to have the gonadectomy was that, in their eyes, this would remove the remaining 'male' parts and therefore make their child more female.

I mean he said ... when he was explaining to us what the situation was, he was saying 'She is a little girl' so we were, like, basically thinking 'She's a little girl, therefore remove the testes.' (P10)

Ironically, where parents felt that they were allowed to be fully involved in decision making, they often found it incredibly hard to decide which course of action would be for the best. They worried that they might make the wrong decision, mainly from the point of view of their child who might later accuse them of having failed them.

But then we start reading where they say that having the ... erm ... testes removed ... some say 'Yes' and some say 'No'. Well once they're gone ... if they say 'No' you don't ... what do you do? It's done. (P1)

(C) The Implications of the Diagnosis for the Child

(5) ISSUES OF GENDER AND SEXUALITY

Given that AIS is a condition in which the sex chromosomes and gonads are incongruent with the sex of rearing, it is perhaps not surprising that many of the parents felt that the condition might have an impact upon their child's gender and sexuality. Often parents reported that they hoped that their child would 'be normal' in every way and this commonly included having the physical appearance, gender identity and sexual preference of a typical girl or woman.

(i) Physical Appearance

Parents referred to a number of aspects of their child's physical development that they felt may have been affected by AIS. An example of this was their child's height with parents being concerned that their children would be taller than average.

I suppose my main concerns were that she wouldn't, in inverted commas, 'be normal' and I saw very much the ... the tall stature and really the ... quite, sort of, big build as ... as actually being an outward sign of ... of not being normal. (P8)

Parents also worried about whether their child would look feminine and have a female body shape with some parents worrying that their child might look more like a boy.

And even when you look at her, you just look at her appearance, you know, whether she's going to be like a boy-looking girl, you know, that's what worried me. (P15)

However, parents also described how, over time, they had become more convinced of their child's femininity. It seems that as their child had developed quite obviously as a 'perfectly normal girl' (P15), their fears of her looking 'abnormal' had dissipated.

But if you saw ten children ... ten girls, her being one, when you've no idea who is ... you certainly wouldn't think she is anything but a girl. (P18)

Although parents stressed how ‘normal’ their children were in terms of their general physical appearance, they also acknowledged that internally (physically) the girls were not so typical. Usually, such comments referred to the child’s lack of female internal reproductive organs or the absence of a vagina.

But what they said was that she was ... to all intents and purposes, on the outside ... a female. and that’s the way it should stay. (P18)

(ii) Behaviour

Although parents acknowledged that it is not uncommon for some girls to be ‘tomboyish’, the knowledge that their child has AIS appeared to heighten their awareness of such behaviour. The parents of children with partial AIS were more likely to raise this as being an issue, although parents of children with complete AIS also described being sensitive to such behaviour.

Some of it stays with me ... erm ... because of the way (daughter) is. She’s very ... erm ... very boyish! She likes boys’ things. She likes boys to play with. She likes boys’ toys, you know, things like that. (P7)

Other parents talked about how reassured they had been by their child’s very female or ‘girlie’ behaviour.

The little one is so girly. The older one is quite sporty but she's still very girly. The little one is ... she's just such a princess! She's just into dressing up. She's much more into Barbie than the other one. She just loves ... she's gone off to my friend's now with four princessy outfits so ... and with her necklace on and her perfume on (laughs). and she's just a real little girl. (P4)

(iii) Gender Identity

For some parents, the information that their child had 'male' chromosomes initially caused them some confusion over whether their child could really be considered female, particularly for the parents of children diagnosed with partial AIS. They acknowledged their concerns about raising a child with XY chromosomes as a girl although, as before, these worries did not appear to last much beyond the immediate days and weeks around the time of diagnosis.

And then when we were told 'We're going to carry on letting her be a girl because she's been brought up as a girl as eighteen months old' and I kept thinking 'Are we doing the right thing? Is (daughter) really a male in a female's body?' (P9)

Many parents described feeling relieved when their daughter appeared to grow up to see herself as female and to identify with her female friends.

What I thought was 'Well they were born as girls and they are girls and ... erm ... they believe they are girls and therefore, there's no two ways about it ... err ... so there's nothing, you know ... I mean it is an issue but, you know, I think it goes quicker than you think (P20)

(iv) Sexuality

Many of the parents interviewed, regardless of the age of their children or their diagnosis, reported that they had previously or still wondered whether their child may be sexually attracted to females as a result of having the male sex chromosomes.

One thing I ... often crosses my mind is when she grows up and starts looking for a partner, what sex will that partner be that she's looking for? Because she's ... obviously a male with female hormones ... what effect does that have? and that's something that nobody can tell me. (P19)

However, most parents reported being reassured of their child's 'heterosexuality' as a woman through the behavioural and emotional evidence they observed as she grew up.

It just used to come to me 'What the hell is going to happen here?' you know, and thank God they are very much into boys so it's a pretty normal thing. I'm so happy. (P20)

(6) THE FUTURE IMPACT OF THE DIAGNOSIS

A common experience of parents who received a diagnosis of AIS for their child was to be immediately thrown forward into thinking quite concretely about their child's distant future. They found themselves thinking much further ahead than most new parents might and, as one parents put it; *The future was unfolding ... we were thinking about it all the time. (P18).*

This category therefore considers some of parents' specific hopes and fears for their child once they had the knowledge that their child had AIS. Fears typically centred on the idea of sexual relationships and marriage but also included concerns that their child would be restricted in the choices they could make regarding careers and sporting activities and would have further painful surgical procedures to go through.

(i) Expecting trauma

There was an expectation for parents that their child's future would be a difficult time. Some felt that the hardest years were still to come and worried about how their child would cope psychologically. Although parents often commented on their child's positive qualities and strengths, they felt it was likely that their future would be an upsetting time for them and some were concerned that this would almost be too much for them to cope with.

I think for the future, you know, 'How we gonna handle this?' There's going to be things that come up and they're going to be upset and ... it's going to be a traumatic time. (P3)

An overriding hope for the majority of parents was that their child would simply 'be happy' despite the many problems they knew they would have to face.

I just want them to be happy ... you know, that they don't have to think about these things. You know, that they don't have to struggle, in other words. (P17)

(ii) Relationships

Some parents wondered whether their children may turn out to be gay and not want relationships with men and thus talked generally about their child's chances of finding a partner. Others, spoke specifically about their hopes for marriage for their child. All those who raised this issue as a concern, hoped that their child would find a partner in later life and indicated that this would be important for their future happiness.

Yeah. I just want them to do what they want to do, have the relationships they want to have, like everybody. (P2)

However, parents stated that their child would need to tell their future partner about having AIS and felt that the child might find it difficult to do this.

Well you also obviously worry that anybody will want them because, you know, you can't go into a relationship and not tell somebody and live a lie. Because that would be very damaging. (P4)

Parents also worried that, as the result of disclosure to a partner, their child might be rejected for having AIS.

So she could be a very unhappy, lonely lady in later life because ... the one person she loves might not want her because of those reasons. (P18)

Their child's infertility also weighed on their minds when thinking about future relationships and parents worried that once a potential partner discovered that there would be no children conceived within the relationship, that they might then not want to be with them.

Ooh, I used to worry whether she would get married, you know, because I think, you know, if people found out she couldn't have children, no-one would want to marry her and that sort of thing. (P9)

(iii) Sex life

Parents were concerned about the possibilities for their child's future sex life and hoped that their child would indeed be able to experience this aspect of life. Some interviewees were concerned about their child's ability to experience the same physical sensations of intercourse as other women and to be able to reach orgasm. Others were worried that it might simply not be possible physically.

I mean intercourse; sexual intercourse is a big part of life isn't it? and without that I think she'd find it very very hard. (P6)

There were also specific concerns about the child's vaginal length and the possibility that this would not allow sexual intercourse without surgery or other treatment.

So it makes you think, well, you know, what sort of operations are they going to have to go through? Are they going to have to spend their teenage years using dilators and that sort of thing and ... erm ... you can almost see your daughter reaching that stage pretty soon and you can see how sensitive they become and you can see what effect it could have on them ... later on. (P5)

(iv) Restrictions on Sporting Career

This aspect of the future was prominent for a number of parents, particularly those whose children were involved in sports clubs or showed talents in this area. These

parents expressed concerns that having AIS would prevent the child from competing as a woman in professional sporting events. They considered how they might deal with this and predicted great unhappiness for their child if she were to be restricted from taking part in the activities she enjoyed and did well at.

I mean ... the reason I was worried ... I can remember hearing stories about, for instance, certain Russian Olympic females being involved in sport and then being disqualified because they had AIS or whatever. (P10)

CENTRAL CATEGORY 1

Powerlessness

'I'll be honest with you - at the moment we parents are lost. That's the fact.' (P17)

The above quote illustrates what appears to be the central category of this section. In talking about their experiences of having one or more children with AIS, the overriding feeling expressed in every category seemed to be one of powerlessness. Parents received the diagnosis 'out of the blue' and were immediately flung into the unknown, feeling guilty for what had happened yet unable to change anything. The lack of information and support appeared only to compound their isolation and feelings of helplessness. In considering their own and their children's futures parents were convinced that there would be difficult times ahead that they were powerless to change. They felt they could have little influence over the treatment their child received from the world and could only hope that their child would develop the inner strength to cope.

It is perhaps within the medical system that the full impact of this central theme is seen. Parents talked about being 'guinea pigs' for the professionals and described how decisions were made for them and advances in medical science were made at their expense. The 'good doctors' were those who listened to parents and gave them information and thus permission to be involved in their child's care. However, the 'bad doctors' ignored them, withheld information from them and let them down. Parents often perceived a need to fight for what they believed their child needed,

although their own needs often remained unmet. Feelings of powerlessness prevailed when parents described their experiences, whether this be reflecting on the past, talking about the present and thinking on into the future.

Table 2

The Disclosure Issue (Child and Others)

PROTECTION					
Central Category	<i>'We're trying to protect them but at the same time we're trying to say, you know, it's not a problem' (P3)</i>				
Classification	Before Telling		The Practicalities of Telling		After Telling
Researcher categories	(1) Life Before Disclosure	(2) Deciding Whether to Tell Child	(3) Deciding Whether to Tell Others	(4) When How and What To Tell The Child?	(5) Outcomes of Telling
Member categories	i. Innocence In Ignorance	i. Reasons Not To Tell	i. Reasons Not To Tell	i. When To Tell	i. Child
	ii. Living a Lie	ii. Reasons To Tell	ii. Reasons To Tell	ii. How To Tell	ii. Others
	iii. Keeping It From The Child			iii. What To Tell	
	iv. Who Knows?				

SECTION 2 - THE DISCLOSURE ISSUE

(A) Before Telling

(1) LIFE BEFORE DISCLOSURE

Around a third of the parents at the time of the study had not yet told their child any details of the condition at all. Others had told their child some but not all of the information about their diagnosis (for instance that they could not have children of their own) and only four of the parents had told their children all of the details of the condition, including the chromosome pattern. All those interviewed, however, were able to remember clearly what it had been like to live with the knowledge of AIS without having told anyone, including the child, about the condition.

(i) Innocence in Ignorance

Parents described a time before the child had any awareness of their condition. They felt that not knowing about AIS allowed their child to remain ‘innocent’ with the implication being that the disclosure of AIS would take away this security and freedom from care.

I hardly ever kiss them or go and see them when they’re asleep because I hate to see them asleep and so innocent and so secure in their innocence. (P4)

(ii) Living a Lie

Many parents gave this description of life before disclosure. They talked about the difficulties of keeping the condition secret, the anxiety it caused them when people enquired over the reason for hospital appointments and treatment and the emotional impact of constantly lying to both their child and others. Although they had their reasons for the secrecy and untruths, parents clearly found this period quite difficult to cope with.

Bearing in mind, all along we've cocooned it because of the fact that it is (daughter)'s business ... therefore, even if we wanted to talk to anyone outside the family, we wouldn't because ... they don't know. (P18)

In many cases, parents had chosen not to tell anyone about their child's condition. However, it was not usually possible to keep their child's hospital appointments and surgery a complete secret and so many had chosen simply to talk about the hernia as a reason for this.

That was what we told everybody when she first had to go up to (hospital). Everybody was wondering why our baby had to go to (hospital), what was the matter with it? So we just said ... erm ... 'The baby's got a hernia. It's got to be operated on'. (P6)

Others found alternative reasons to explain the trips to hospital. Some avoided mentioning hospital altogether, others invented less complex medical reasons or those which they felt able to discuss without feeling so awkward.

I think we've just sort of told white lies about coming. Having treatment for asthma or, you know ... (P8)

One aspect that parents did often feel prepared to share with others was the reality of their child's infertility. To some extent, this seemed related to the fact that they were likely to discuss this with the child themselves at quite a young age and similar explanations appeared to be given to other people.

I think I'd told people she can't have children - that ... erm ... she doesn't have a womb. But I haven't told them the full truth. (P7)

(iii) Keeping It from The Child

In trying to keep things from their children, some parents reported that they had tried 'fobbing' their child off or even lying to them to prevent them from learning about their condition.

We've consciously ... we've never ... I've always ... I've always felt it's wrong to lie but there are ... you can twist things slightly. (P18)

However, many reported that their children had begun asking more and more questions and that it become increasingly hard to avoid the issue.

and that was very difficult because they were ... very articulate and very aware of everything and ... inquisitive ... and that was hard. (P1)

(iv) Who Knows?

Those parents who had chosen not to disclose their child's condition to anyone described feeling deeply concerned that other people might find out. They expressed a belief that it was almost inevitable that the information would 'get out' (P18) and the idea of not knowing who knew was a source of great anxiety for some.

I want to know who knows. I don't want it to seep out in a way that I ... somebody knows and I don't know of it. You know, like gossip starts. (P4)

Parents described some of the specific routes via which other people might discover their child had AIS and expressed a concern that the confidentiality promised by the medical system was not as water-tight as it should be.

and ... erm ... the other thing, we went to (local hospital) and there were people ... the doctors and ... people that we socialise with but who we wouldn't want to know our business. Well the secretaries - they'd been through it [the notes] so they knew. (P18)

(2) DECIDING WHETHER TO TELL CHILD

All the parents interviewed stated that they felt that their child should know that they had AIS and the implications of this, although few of them had actually made a full disclosure. They described their thought processes in reaching this decision and gave a variety of reasons to support their thinking.

(i) Reasons Not To Tell the Child

The parents of older children (i.e. from teens upwards) commonly reported that the consultant who had given the diagnosis and who had provided follow-up in the early years, had advised them against disclosure to their child. Although some parents disagreed with this view, they reported that the recommendation of a medical consultant had been something that they'd found difficult to dismiss.

(Doctor) initially told us that whatever we do, we must never tell our daughters because it would be psychologically damaging for them to realise that they were biologically male. (P13)

Parents also had quite considerable concerns about the way in which their child might react to the diagnosis. This ranged from being worried that their child might be upset, to being afraid that they would be thrown into a state of extreme psychological distress and even experience a complete personality change.

Psychologically I worry about how she might be psychologically affected. Whether she'll become severely depressed or suicidal and ... erm ... stuff like that. (P1)

There were worries that the news would cause the child to doubt their identity. Parents suggested that they might feel different compared to their peers or feel that they were 'weird' in some way.

I think that's my biggest fear. That they're not going to think of themselves in the same way, you know ... as we think of them really. (P3)

In addition to the concerns about the children's identity in general, parents were also worried that the diagnosis might trigger a gender identity crisis.

I mean, you don't want a child of six having a sort of gender identity crisis about what ... err ... what their gender is. (P8)

Another reason given for non-disclosure was a fear that the information about AIS might cause the child to become angry with their parents. There were concerns that the children might blame their parents for giving them the condition or that they might be angry that the right treatments had not been chosen. Parents expressed their fears that the bond with their child might be disrupted by the disclosure.

I thought ... you know, 'I'm never going to be able to tell her' and, you know, 'Is she gonna hate me for it?' or blame us for the condition that she's got? You know, it's hard. (P6)

(ii) Reasons to Tell the Child

The most commonly cited opinion by parents concerning the disclosure to their child was that she had 'the right to know'. Whatever their fears and misgivings about actually telling their daughter about her condition, the parents were clear that their child should know ... whenever that might be.

I feel she has the right to know exactly what I know. It's her body; she's got to live through it. It's her life. (P9)

Parents with younger children had generally been told by the doctors that disclosure should be made and those with older children often found that the consultants who had previously advised non disclosure had later changed their minds or new doctors with different opinions had replaced them.

He's telling us to be open and to explain ... you know ... what's what with (daughter) and not to hide things (P1)

Parents were often aware of stories of individuals with AIS who hadn't been told of their condition and who had found out in later life. The pain and upset that they believed this type of discovery to have caused was enough to make parents believe that their own children must know about their condition at a relatively young age.

But I mean I think a lot of it applies to adults who didn't find out until ... you know, didn't find out about their condition until they were older and then actually ... erm ... were sort of obviously very angry that the whole thing was sort of shrouded in secrecy. (P9)

A number of parents were also concerned that without disclosure, their children would worry that their trips to hospital were actually a sign that something was seriously wrong. These parents felt that this was a reason to tell them the truth of their condition.

and when we came out of the room, (daughter)'s face ... because she'd been sent out ... and it was 'What did he say? He was talking about me wasn't he? I know there's something really bad with me!' and all the picture ... all the questions that were going on in her mind was she thought that she had something very very seriously wrong with her health ... and the doctors weren't telling her. So that was a fault of ours. (P6)

(3) DECIDING WHETHER TO TELL OTHERS

Many parents in the study, even those who had made a full disclosure about AIS to their children, reported that they had told very few other people about the condition. Other parents had chosen a select few other people to talk to. Reasons for these choices are given within this category and the outcomes of such disclosure will be described later.

(i) Reasons Not To Tell Others

The major reason given for non disclosure to others was a feeling that the condition was the child's business. Parents believed that until their daughter was old enough to know about the diagnosis and to make the decision for herself about who should be told, they did not have the right to tell others without her consent. For this reason, despite an often stated wish to 'get it off their chest', parents chose to hold off any form of disclosure to outsiders.

and very early on we decided that ... we talked about the aspects of sharing this with other people and decided, for the girls' benefit, we wouldn't put word around ... about it and then they could decide in older age whether they wanted their friends to know. (P12)

Parents were also concerned that once others knew about their child's condition, that they might be unable to help themselves from seeing her in a different light and thus treating her differently. They didn't wish their child to be singled out and seen as different because of her condition and so often chose not to disclose.

I just think that they're ... they're entitled to privacy. I mean they are going to ... umm ... stand out. It's different, it's something that's really different and it's something people will talk about. (P2)

Following on from the above idea, was parents' belief that, not only might their children be perceived as 'different', but that they might then be picked on or teased because of their condition. For most, this concern was for their child's school days although for some parents the idea that their child might be persecuted in adulthood too was also an issue.

It could cause problems at later dates. There's bullying in school. That sort of ... kids find out and all that sort of thing. So there was those worries ... (P10)

Other reasons that parents gave for not telling, related to their concerns about how the information would be taken. Parents worried that others wouldn't be able to understand the diagnosis or that they might worry once they knew. Non disclosure to others was therefore a way of sparing themselves from the difficulty of explaining, and others the difficulty in understanding and coping with the information.

You can't just explain to people, I mean it's a condition where they ... people ... they don't know and then they misunderstand completely. (P15)

I was very very tempted, I must admit, to tell him but I just didn't have the heart to do it because it would have devastated him. I don't think ... because they were a different generation and I don't think he would have taken it well at all. (P14)

(ii) Reasons to Tell Others

Often it seemed that where others knew about the condition, this was through necessity or accident rather than through a conscious decision about who to tell. For instance, where immediate family were aware of the child having AIS, this was generally because grandparents had been present at the diagnosis, or parents had been so shocked and upset by the news that they had needed to immediately talk about it with their closest family or friend.

I needed them [interviewee's parents] there then. I needed them. I didn't need extended family then, I didn't want other people to know but I wanted them. I needed help to get through it. (P7)

Almost all the parents talked about needing to tell someone else, even if it was just one person other than their partner. For most, the choice had been to disclose to their own parents - if not at the time of the diagnosis, then later.

I mean we ... err ... discussed it actually with ... erm ... at home actually with my ... my parents were there. My brother was at home. and later on I think (wife) discussed it with her parents. (P20)

For some, however, having disclosed about their child's condition to friends at this stage, they later reported regretting having done so.

and I think, 'Oh, I should have kept my mouth shut. I shouldn't have told that person.' At the time I had ... when I was going through it I had a really close friend and I ... I told her but now we're not so close - not that we're not friends anymore but we're not as close now as we were then and I think 'I wish I'd never told her that.' (P7)

It was emphasised that the people whom parents chose to disclose to were carefully chosen as being people who could be trusted not to pass on the information to anyone else.

I mean they were people I could trust not to discuss it with other people (P8)

Some parents explained that they had chosen whom to talk to about their child's condition on a 'need to know' basis. For some this meant telling a close relative so that if anything happened to them as parents, that someone else would be aware of the need to continue treatment etc. For others it was an issue of the inheritance of the condition and the possibility that other family members might have the AIS or carry

it, thus meaning that they might pass it onto their children without having any awareness of it. Parents also outlined situations where they felt that other people definitely did not need to know such as their child's school.

Because (wife)'s obviously a carrier, we ... you know ... then had worries about (wife)'s sister and her little girl and what not as well so we had to tell them really. It was only fair to sort of tell them. (P3)

I thought 'Aha, what happens if something happens and there's nobody around?' and I told my sister. I think I just said 'You know (daughter)'s got this thing and it is ...' What was very important was that she had her testes removed because they can turn cancerous and it hadn't been done. (P13)

(B) The Practicalities of Telling
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(4) WHEN, HOW AND WHAT TO TELL THE CHILD?

Parents in the study were at quite different stages with what they had chosen to tell their child and when and how they had gone about doing that. As we have seen in the method chapter, the children ranged in age from 3 to 21 years (median age 11½) and only four interviewees (including the two with the oldest children) had given their daughters complete details of the condition including the information about their chromosomes. Parents often had quite strong feelings about what their child should and should not know about the condition and at what stage.

However, it often seemed that their personal feelings about the difficulties of giving such information and the fear of the diagnosis being further disclosed were as much involved in such decisions as the impact they felt that it might have on the child.

(i) When to Tell

Parents rarely had a specific age in mind at which they felt their child should know particular pieces of information although almost all talked about particular triggers to act such as the need to have an explanation to give to their child when hormone replacement treatment was commenced (usually from around the age of 10) or when she might be expecting to begin her periods.

In fact, many of the parents, particularly those with younger children, stated that they had 'no idea' about when to make the disclosure although they often talked about a growing awareness (and sometimes fear) that the disclosure must be made 'soon'.

I suppose I have to start telling her bits soon. (P4)

Others admitted that despite being sure they wanted their daughter to know about her condition, they were 'putting off' the event because they were uncertain about how to go about it.

I think we're both quite reluctant to sort of, you know, take our feet off the brake and move onto the next stage ... the next step. (P5)

Although parents may not have had a particular age in mind by which to make the disclosure, the main consideration in deciding when to give information, appeared to be their perception of their child's maturity.

I don't think they have the right to know at an early age because I think it's too ... too complicated an illness to take in and understand. (P9)

Some parents stated that they would only know that their child was ready to have the information when they began asking questions about their situation. Although this reason was often stated as a strategy to ensure disclosure at the appropriate time, it could also be seen as a way of avoiding broaching a difficult subject. This strategy is at odds with the view presented in the category of 'Reasons to Tell Others' where parents acknowledged that children might worry that something more serious was wrong with them but not want to ask questions about this.

and she can talk about things. I don't think she's bottling things up. I think that when she really needs to know, she will ask. (P1)

In the category 'Reasons Not To Tell Others', most of the parents were clear that whenever the disclosure were to be made, their child would always receive the information before other people did and that it would then be up to them to decide

who else they wished to know. However, the interviews revealed a dilemma for parents when they reached a time where they felt their child had the maturity to understand and cope with the knowledge of their diagnosis, but not necessarily the maturity to appreciate the consequences of telling other people the information. This was a key factor for many in deciding when to tell their daughter about AIS.

I don't know when she should know because children talk amongst themselves and I think she's not going to know ... the right person to tell or not to tell or ... you know.

(P7)

(ii) How to Tell

Many parents stated that they simply had no idea how to go about telling their daughter about AIS.

and that's why I sort of said to (doctor), you know, 'I need help. I need someone to talk to because I haven't got a clue how to tell her.' (P11)

For some the concerns about how to tell appeared to be related to a fear that they might 'say the wrong thing' whilst for others it was a fear that they would not have the answers that their child needed. Parents who felt that they did not fully understand the diagnosis were more worried about how they would explain it to their child.

I'm so worried about getting it exactly perfect that I'm scared to do anything. (P4)

The only real strategy that parents were able to describe of how they had told or planned to tell their children about the condition could be described as the 'bit by bit' approach. They stated that the disclosure should be spontaneous rather than formally arranged and that it would be better for the children to gradually come to learn the truth over a period of time.

The way in which (child's mother) is dealing with it is letting her know little bits slowly at a time. (P10)

In addition to their thoughts about how they would actually physically tell their children about the condition, many parents described how they had tried to ensure that when they made the disclosure, they would have resources to support this. They were aware that simply hearing the news from their parents may not be sufficient for the child and so they had gathered together information to give when the time arose. Parents also talked about setting up support systems for their child so that there would be people other than their parents whom they could talk to when they learnt about their condition.

and I kept a magazine clipping, you know, 'Take That' magazine. There was an article on (daughter)'s illness from a girl who was ... I think she was about eighteen or nineteen ... erm ... I kept it and I thought 'I'm going to show her that.' (P9)

She may immediately react towards me and (mother). and blame us as her parents so therefore she's then going to look for ... comfort with other people so ... I think the more people around her personally, I mean family and friends that know ... and the more information they have, the better it is. (P10)

(iii) What To Tell

It seemed that disclosure, for parents, was never a one-off event. Rather, there were a number of stages to the information giving process which could be roughly separated according to the degree to which the information related personally to the child. Most parents reported that the first stage involved talking very generally to their child about the ways in which people could differ from the 'norm'. Often this involved using examples to illustrate how people very often have problems in life. Specifically, this time was used to introduce the idea that not all women are able to have babies.

When she was about six she was talking about how many children she was going to have etc. etc. and I took the opportunity at that time to say 'Well actually, not everybody can have ... err ... can have their own children for various reasons.' We actually had our next door neighbours who had adopted their children so I was sort of able to bring that up. (P8)

Early explanations to their children about the medical investigations and treatments that they had gave parents an opportunity to begin to introduce the idea that their

child's development had not been completely as might perhaps have been expected, without talking about AIS. At this stage, parents explained that there had been parts of her that hadn't been finished off properly and that had sometimes required medical intervention.

We said that when she was ... erm ... in the womb she didn't develop properly. (P18)

Additionally, parents explained about surgery, particularly the gonadectomy, by telling their children that they had had parts which were 'poorly' or shouldn't be there and that had been removed by the doctors.

I think too, we must have said to her that 'You've got some bits that aren't ... some nasty bits that need to be taken away.' (P1)

Having spoken generally about women who couldn't have their own children and having given the information that something had been wrong when the child was born, parents had then been able to introduce the idea to their child that she had no womb (or sometimes ovaries) and that she wouldn't be able to bear children.

I told her she was born without a womb you know ...and without ovaries so she sort of wouldn't be able to have children ... (P6)

Having explained some of the basic implications of AIS to their child (although usually without telling them fully about the condition or giving it a name), those parents whose children were old enough to need hormone replacement treatment had been able to account for it by following on from these simple explanations.

Just sort of saying to her that because she hasn't got any ovaries, that's what produces your hormones so they are going to give you some tablets to make your hormones. (P6)

The next stage for parents appeared to be to give the child the full information about her condition including it's name and this seemed to be a stage that took parents a relatively long time to begin. of all the children represented in this study, only two had a full knowledge of their condition and even for these, the parents had chosen not to give the information about the chromosomes at this time. Only one other child was aware of her chromosome makeup but she had not been told about having had testes that were removed. There were concerns about divulging this type information to their child for reasons that have been covered partly in the category on 'Reasons Not to Tell the Child' and parents seemed generally to believe that this information should come much later in life.

We've never actually said to (daughter) that she's got XY chromosomes ... I think that's about the only part that ... because that should come in later life, I think. (P6)

(C) After Telling

(5) OUTCOMES OF TELLING

(i) The Child

Although only around a third knew everything about their condition including their chromosome pattern, at least two thirds of the children had been told some aspect of their condition - usually that they would not be able to have children. The parents talked about how their children had reacted to the information they had given them and more positively, a number of parents commented on how quickly and easily their child had understood the information and how well they had coped with it.

My main concern was about how she was going to react to the fact that she couldn't have children but to be quite honest, she took it ... really well. (P9)

Earlier categories have described how parents were concerned that knowing about AIS might cause the child great psychological distress or result in them becoming angry with their parents. They also worried that the disclosure might cause the child to doubt who they were or even trigger a gender identity crisis. For most of the parents, however, the disclosure did not result in the extreme negative outcomes they were expecting. For example, although children were sometimes upset at the news that they couldn't have children they were not hysterically so.

She knew at the age of nine that she wouldn't be able to have children which ... that was a big shock to her at first, even at the age of nine when you don't think children sort of think about having children. That came as a sort of ... that was upsetting.
(P6)

The children sometimes reported to their parents that they felt different in some way when they knew about the consequences of their condition. The fact that they would not have periods was one such aspect, although sometimes - particularly when mothers had presented this as a positive aspect of the condition - the children had reported feeling quite pleased about this!

I think now she's becoming more ... not emotional but ... she's aware that her friends are starting their periods and things and she said to me 'I'm different, I'm not going to have periods.' (P8)

Only one parent reported that her child had been concerned about getting a partner as a result of not being able to have children, another child had admitted to worrying that she would be a lesbian and three of the parents reported that their child had made comments about her gender identity. In all of these cases, it appears that these worries had been reduced through parental reassurance and, according to parental report, had not developed further.

and I said to her 'What's the matter?' and she said 'I'm a boy!' and I said 'Sorry?' and she said 'I'm a boy. If you look in that book, he hasn't got any ovaries.' (P11)

Despite their concerns about the impact of the disclosure, a positive aspect of the process that parents talked about was the sense of relief they experienced or expected to experience through telling their child about the condition.

It's like unburdening it when they're in control of it, when they know. (P4)

Only one of the children who had received the full details of her condition was reported to have been 'devastated' by the diagnosis and in this case, her younger sister had reacted very well to the same disclosure. It seems that this one child may have had additional problems which made it more difficult for her to come to terms with the diagnosis of AIS and its implications.

(ii) Telling Others

Parents had naturally been cautious about telling others about their child's condition (usually because they perceived it to be the child's business) and for many, as we have already seen, this meant telling only immediate family. On the whole, the experience of telling these select people had generally resulted in reactions that the parents found acceptable.

A main concern before telling others about the condition was that they would be

upset by the news and this had sometimes been the case.

Our parents were very upset. I think with parents, and particularly my parents, it just made them realise ... erm ... what you've gone through. (P5)

However, many of those parents who had made a disclosure to someone else reported that the reaction they had received had generally been very supportive and that they had found this helpful.

I remember my sister-in-law ... err ... she told me that 'It's OK' she said 'So what? They all said the same thing, 'So what if they ... so what if she can't have any kids? You know, she can adopt.' (P17)

Disclosing the condition to another person was something that many parents experienced as a great relief. In particular, talking to a close friend was something they found to be very helpful.

I think just the process of actually telling someone ... you know, what's going on, you feel it's a sense of relief, you know, it's not just your own thing anymore. (P7)

A few parents had experienced reactions from others that they had found less helpful. It seemed that the reaction that parents most commonly reported as being unhelpful was that of other people downplaying the news with such comments as 'It could be worse.' Although parents frequently used this type of thinking themselves in coping

with the condition, they perceived this reaction in others as implying that they didn't really understand quite how big an impact it had had upon them as parents.

My mum tries to help a lot but like 'There's a lot of children that are worse off and at least there're not going to die'. and I feel like saying 'No, but that doesn't mean I want anything to be wrong with mine.' (P2)

In some cases, parents were upset by the reaction of medical professionals who had been told about their child's condition.

and we went in ... and he sort of said, 'Oh, is this (younger daughter)?' Oh my god, she's got it too has she?' (P2)

CENTRAL CATEGORY 2

Protection

'We're trying to protect them but at the same time we're trying to say, you know, it's not a problem' (P3)

The theme of protection seemed very much to sum up parents' experience of the disclosure issue. The dilemma described by the parent in the above quote describes the position of the group as a whole in thinking about telling their children and others about AIS. Their abiding feeling was one of wanting to protect their children from harm and it was difficult for them to know which would cause more harm - to tell or not to tell. By telling their children, the parents perceived that they might cause untold damage but by not telling them, they feared that their child might suffer greater upset in the future.

By not telling others about the condition, parents hoped to spare their child from being treated differently, even cruelly. Non disclosure to others might also be seen as protecting the parents to some extent, by sparing them from the difficult reactions they might encounter. It was also seen as being protective towards those who might receive the news and then also be burdened by it. However, by not telling others, the condition was kept secret and, as one mother put it, life was made to seem 'seedy'.

Parents were very clear that their children were 'normal' but that their desire to protect them meant that they themselves were further perpetuating the view that

children with AIS are abnormal. The need to protect appeared to be more powerful, however, than the desire to change attitudes and appeared to be the issue underlying their thoughts and actions about disclosure.

CHAPTER 4 - DISCUSSION

Introduction

The experience for parents of having a child with androgen insensitivity syndrome (or any other intersex condition) is a much under-researched area. The current study aimed to discover what the experience is for parents following the diagnosis of AIS for their child and to understand better the issues that affect parents' decisions about whether, when, how and what to disclose to their children and others about the condition. A grounded theory methodology was employed to provide a detailed picture of participants' experience and hence to offer insight into complex real life phenomena (Strauss, 1987).

Due to the large volume of data collected and the many categories represented, this chapter will focus upon the most significant points arising in the analysis. These major findings will be discussed with reference to the existing literature. A consideration of methodological issues will be followed by a discussion of the limitations of the current research and implications for future studies. The final section of the chapter will address the implications of this study for clinical services and for clinical psychology practice.

Receiving the Diagnosis

In considering the reaction of parents to the diagnosis, Slijper *et al.* (2000) found that the majority of parents reacted with shock. The current study found that parents receiving a diagnosis of AIS for their child experienced it as ‘a bolt from the blue’. The shock of the diagnosis was then seen to affect their ability to ‘take in’, process and understand the information they were given. This moment in time was also very much characterised by emotions of devastation, disbelief, denial and confusion.

For many parents, their recollections of what they were told at the time of diagnosis were limited to a few words or phrases. It is not known what facts parents were actually given during the consultation, but it seems that the shock of receiving the diagnosis often prevented parents from retaining much of the information with which they are presented. In the research interviews it became clear that for some parents, this was the only information they received about the diagnosis and even several years on, some appeared no better informed about AIS than they were at the time of the diagnosis.

Slijper *et al.* (2000) found that receiving full information about AIS when the clinical diagnosis was made was one of the most important factors in determining parental coping. However, it seems from the current study that whatever the facts that are presented to the parents, what they actually took away was not the full information.

The Later Emotional Impact

As the initial shock of the diagnosis began to wear off, it seems that other strong emotions took its place. Slijper *et al.* (2000) described an emotion of anger in parents following the diagnosis of AIS. However, 'anger' was not an emotion that was commonly expressed or referred to by parents in the current study. Where it did occur, it was in relation to parents' experience of the medical system and arose more in terms of frustration at the way in which they were dealt with by professionals.

Grief was also an emotion that Slijper *et al.* (2000) identified. However, because their study involved ratings rather than qualitative analysis, they were not able to directly link these feelings with specific consequences of the condition. In the current study, grief was seen to occur for almost every participant and this was usually directly related to the discovery of the child's infertility. This phenomenon can be conceived of as a process of mourning for the loss of their child's expected future children and the parents' future grandchildren. A sense of grief for the future also occurred for many parents who feared that their children might be deprived of having satisfactory futures including relationships and sexual intimacy. This idea of bereavement was one that many parents could associate with and one which was still very prominent for the majority at the time of the interviews.

Parents in the current study also experienced feelings of guilt and blame. Early in the research, the interview schedule did not include a specific question about this aspect of parents' experience and neither did parents spontaneously mention this. However,

when a general question was asked about whether parents had ever experienced such feelings, almost all of the mothers (rather than fathers) responded that they had indeed felt some sense of guilt for their child's situation. This emotion was related to their belief (whether confirmed medically or not) that they had passed this condition onto their child either genetically or because of a problem during pregnancy.

The Support Parents Want

A third of the parents in Slijper *et al.*'s (2000) study reported having received psychological support at the time of diagnosis whereas the majority of parents in the current study reported that they were not offered any type of support at all when they were first informed about the condition. However, most parents also reported that, had counselling been offered, they would not have taken it up. Parents generally did not identify this as a need in the early days but expressed, rather, a need to meet with others in a similar situation to themselves. They wanted to talk to other parents who had already 'been through it' and who were perceived as being more likely to be able to understand the issues they were facing and to offer relevant support and information.

Parents reported that the existing AIS support group, whilst having a large membership of adults with AIS, did not meet many of their needs. When the diagnosis was new to them, it was often too hard for them to hear the issues that adults with AIS appeared to bring to the meetings concerning their surgical treatment and gender assignment.

For the parents in the current study, it seemed that the support group often caused them to feel more anxious and worried about their children's future and more convinced of the difficulties their children will face in the future.

Parents who had been to meetings or spoken to members of the group often reported feeling overwhelmed by the strength of emotion and opinion expressed and often didn't return.

Disempowerment by the Medical system

This phenomena of disempowerment and helplessness within the medical and professional system is one that appears not to have been investigated previously in relation to intersex conditions, yet was important enough in the current study to be seen as the central theme pervading the whole of parents' experience. It would seem appropriate to consider the way in which parents experience the systems that they become involved with as a result of their child's diagnosis.

Not every parent's experience of the medical system was perceived as negative, although all the parents spontaneously gave examples of less positive experiences, even if this were simply a complaint that a local hospital or GP displayed a lack of knowledge or sensitivity in dealing with them and their child. However, more negative comments were made than positive and it appeared that for some parents they felt at best ignored and overlooked by those responsible for their child's care and at worst felt let down, insulted and even disliked by their doctors.

Many parents felt that they did not possess the relevant knowledge or status to be in a position to take a more active role in their child's treatment, and a common feeling was that they, as parents, had little say in the decisions that were made. Where parents did challenge the system, they were often greeted with resistance and some even felt that they were 'disliked' by their doctors for challenging their wisdom and authority.

Anecdotally, it appeared from discussions with parents, that those who were better educated or of a social position closer to that of their consultant, had a better experience of the medical consultations and were more able to negotiate over the options for their child's care. It may be that the 'one-down' position that parents feel assigned to is one that is fostered by the medical system (perhaps unconsciously) due to their own lack of certainty regarding the treatment and outcomes for children with intersex conditions.

Fears of Psychopathology

Funck-Brentano *et al.* (1997) claimed that parents' primary concern in HIV was to protect their child from depressive or fearful reactions, and this idea came across strongly for the parents in the current study when thinking about disclosure of AIS to the child. One of the main arguments given for non-disclosure in the current study was the fear that it would cause psychological distress in the child.

One of the only studies which focuses on the outcomes for children with AIS is that by Slijper *et al.* (1998). Other studies have considered the outcomes in adulthood for individuals who were raised in a time when a diagnosis of AIS was generally not discussed with children at all. Slijper *et al.*'s (1998) study included a small number of children with CAIS and PAIS but they found that approximately 50% (6 of 12) of the children with CAIS and 13% (1 of 8) of the children with PAIS exhibited general psychopathology (selective mutism, anxiety, sexual problems, oppositional defiant disorder or depression). However, the study does not make it clear whether the child was aware of their diagnosis or not at the time of onset of the problems described.

In the current study only three of the 17 children (17.6%) were reported to be experiencing difficulties in terms of internalising or externalising problems as reflected through their behaviour and reported using the Child Behaviour Checklist. Two of these, however, were above the age cut-off to technically be able to assess using this measure and the third was a child with PAIS who was experiencing significant medical (and thus social) problems as a result of a surgical reconstruction of her vagina when she was younger. It does not seem from the current study that having AIS is necessarily a strong indicator of psychopathology, however, given that two of the girls who did experience significant problems were also two of the only four (i.e. 50%) to have full knowledge of all aspects of their condition, this is something that should be explored further.

Slijper *et al.* (2000) found that psychological problems were twice as prevalent among children who did not receive psychological support and help as soon as the diagnosis was made. They believe that early counselling has a preventative effect, although even adding this element to the package of care families receive does not appear to be sufficient to keep all children from developing signs of psychopathology. The current study cannot comment on this hypothesis, however, it is known that whilst a few of the children had met with a psychologist on one or two occasions, most had not had any contact with services and realistically, are unlikely to come to the attention of psychology unless a problem is identified by their parents or medical consultant.

Gender-Related Concerns

Parental coping with the gender assignment

Children with complete androgen insensitivity syndrome (CAIS) are usually raised as girls (Slijper *et al.*, 2000) and there appears to be little question about the appropriateness of this course of action. In fact, in Slijper *et al.*'s (1998) study it was stated that in cases of CAIS, nearly 80% of parents were comfortable with the assignment to female gender despite the incongruent chromosome pattern. In the current study it seemed that parents of children with CAIS did not even consider that their child had been 'assigned' to a gender but rather that their children were girls and that raising them as such was not an issue.

However, for the parents of the children with partial androgen insensitivity syndrome (PAIS), the decision about gender assignment was much more pertinent since the criteria used to decide the management of infants with PAIS in terms of their sex of rearing is still an area surrounded by inaccuracy and confusion (Viner, 1997). Those parents of children with PAIS in Slijper *et al.*'s (1998) paper were said to have much more difficulty coping with the gender assignment of their child and some 50% of these parents were felt to be unable to deal with the lack of gender clarity - to the extent that three parents openly rejected their child as a result (Slijper *et al.*, 1998).

Only four of the parents interviewed in the current study had children with PAIS and three of these reported that in the early weeks, months and even years, that they had had concerns whether the gender assignment would prove to be the right choice for their child. Even at the time of the interviews, although these parents reported that they had become more and more convinced of their child's female identity over time, they still appeared to be much more aware of their child's 'male' beginnings.

Donahue (1987) reported that if the child's physical appearance is discordant with the chromosomal or expected sex then it is more likely that parents will have difficulties in accepting the child's assigned gender. This may be the case for the parents in the current study; however, the study only included a small number of parents of children with PAIS and did not focus on this particular question.

Gender Identity and Gender Role - how the child sees herself and how she behaves

Although the parents of children with CAIS did not report any worries about their child's sex assignment a number of these, in common with the majority of parents of children with PAIS, did express some concerns over their child's gender identity - the gender that their child perceived herself to be. None of the parents of children with CAIS raised any concern that their child might not see herself as female - although sometimes they were worried that the news of her diagnosis might confuse this. However, most of the parents of children with PAIS revealed that they had been worried about the long term outcomes for their child in terms of gender identity. They were concerned in the early days that their daughter might feel 'male' and be confused by being raised as a girl.

Slijper *et al.* (2000) hypothesised that it is the gender that parents *felt* their child to be which influenced how they dealt with their children and thus, if the external appearance were not completely female at birth, there would be confusion within families about the 'real' gender of the child. Slijper *et al.*, (2000) therefore postulated that for children with AIS, it was the parental attitude to the child's gender that provides one of the most important contributors to a child's gender role identification and gender identity. It seems possible, from the current study, that this might be the case since it was commonly the parents of children who had been born with ambiguous genitalia who raised the most concerns in this area. However, with such a small sample of children with PAIS within the group, such conclusions cannot be made firmly at this stage.

In terms of gender role, Slijper *et al.*, (2000) found that the children with PAIS raised as girls were more likely than the children with CAIS to show masculine gender role behaviour and hypothesised that one reason for this was the increased exposure to pre and post-natal androgens in children with PAIS. In the current study, all but one of the parents of children with PAIS and approximately a quarter to the parents of children with CAIS commented on their child's tomboyish behaviour and made a link between this and AIS. Some wondered if the condition might be responsible for making their child more 'boyish' and others questioned whether their child's behaviour was actually unusual or whether they were simply more sensitive to signs of masculine role behaviour compared to parents of children without AIS.

A question about parental thoughts regarding their child's sexual preference was an area that was incorporated into the interview schedule when it became clear that this was an area of concern for many parents. When asked, most of the parents admitted to having considered the fact that their child might be homosexual in her choice of partner and, again, they often linked this to the fact that she had male chromosomes.

The majority of the parents, however, reported that over time they had become less worried about their child's behaviour and preferences. This was mainly linked to their perceptions that as their daughters had grown older; they had begun to take more of an interest in 'girlie' activities and hobbies. Those parents of children with PAIS appeared more likely to have continuing anxieties in this area, however, no parent described a masculine gender identification in their child or any of the

symptoms of a gender identity disorder or body image problems.

Gender Identity Crisis- how the child reacts to the diagnosis

Parents main concern in the area of gender appeared to be a fear that through the disclosure of the condition, they might trigger a gender identity crisis in their child. They worried that by knowing the details of the condition, that their child's confidence in her own identity would be shaken and that she might find this too difficult to overcome. It is perhaps for this reason that only a few of the parents in the study had actually disclosed to their child about her chromosomal makeup. Although this wasn't explicitly investigated in the study, it is possible that parents felt that this piece of information was the most likely to trigger gender identity problems since it reveals that the child is biologically 'male'.

The literature in the area of gender identity disorder in intersex conditions is conflicting, with authors such as Slijper *et al.* (1998) reporting that children with a physical intersex condition constitute a high risk group as regards the development of disorders of gender identity. Others, however, report that almost all patients assigned to a gender incongruent with their chromosomal sex are content with the gender in which they are raised and very few show a gender identity or gender role in accordance with their chromosomal sex (Migeon and Wisniewski, 1998).

Disclosure to the Child

The parents all stated that they felt that their child should and would be informed about her condition, however, they did not have clear ideas about how this would take place and many commented that they were 'putting it off' since until such circumstances arose that would mean that the need to disclose was more pressing (such as the need to explain HRT or a child's questions about her medical appointments). Parents predicted that both the disclosure and the time immediately afterwards would be hard and it is perhaps not surprising, therefore, that they were often unable to form any clear plan about how they would go about beginning the process.

Goodall (1991) asserted that in any disorder of early onset, including those which involve a chromosomal abnormality, it is better that this is revealed to the child gradually and at appropriate stages of cognitive and physical development. Parents agreed that the age at which a child should know of their condition should largely be defined by that child's age and maturity and that the disclosure should be made gradually over a period of time.

The 'bit by bit' method of disclosure favoured by parents is also that recommended by researchers and clinicians for conditions such as cancer as the best option in the diagnosis of cancer (Spinetta, 1980) with disclosure geared to a child's level of cognitive development and psychosocial maturity (Woodard and Pamies, 1992). Slijper *et al.* (2000) report that before a child with AIS is ready to be informed about

their condition they will first need to receive early education about the biological and psychological aspects of normal sexual development so that they have a baseline from which to be able to understand the ways in which their own body differs. A number of parents in the current study reported that they had had to teach their child basic biology in readiness to tell them about AIS and that they had found this to be a challenge.

In terms of what the literature advises regarding the age at which different parts of the diagnosis should be disclosed, Warne (1997) suggest that between 6 and 11 years children should be given simple explanations for the reason why they need to see the doctor. Then by the age of 11 or 12 years (or before hormone replacement therapy commences all information about AIS should be given with the exception of the XY karyotype such that a girl with AIS is able to understand a discussion about the complex nature of her condition and its implications (Warne, 1997). Nearly two thirds of the children (10 out of 17) in the current study were 12 years or younger and only three of these had the level of knowledge recommended by the above researchers. The majority of children in this age range were only aware, at most, that they had no womb and could not, therefore, bear children.

As regards the disclosure of the chromosome pattern, it is recommended that this occur between the ages of 15 and 17 - before children learn about chromosomes in school (Warne, 1997; Slijper *et al.*, 2000). It may be particularly important for older children to know more about their condition both to help them make decisions about treatment and to help them to explain to others if necessary. of the children aged 15

and over, all but one of these were aware of all aspects of their condition *including* the chromosomal information.

In comparing the disclosure patterns recommended in the literature, with the reality described by parents in the current study, it would appear that many parents may begin the process of disclosure later than is perhaps advised. Whilst parents may find it difficult to talk with their child about the condition, in the cancer literature, it is said that a lack of disclosure may communicate the unintended message that the condition is a morbid, frightening secret to be worried about and that it is so toxic and dangerous that it cannot be discussed openly (Claffin and Barbarin, 1991). Indeed, parents within the current study acknowledged that if their children did not receive information about their condition they may worry that something terrible must be wrong with them.

Overall, it seems that whilst parents are convinced that it is right for their child to know about AIS, they are hesitant to begin this process. Parents talked about their child's 'innocence' before disclosure had taken place and the fear of shattering this with the news of the condition appeared to be one of the main impediments to telling. The parents' fear of causing psychological distress or a gender identity crisis was a major obstacle to the disclosure process. Without information about the outcomes for children with AIS after disclosure, or psychological support to see them through this time, it is perhaps not surprising that many parents did not achieve the recommended 'goals' in terms of the amount of disclosure to be achieved by the time their child was 12 years old.

Methodological Issues

Qualitative methods are said to avoid the simplifications imposed by quantification by allowing individuals to be studied in depth and detail and enabling the more complex aspects of the experience to be studied (Barker *et al.*, 1994). However, although qualitative research is carried out in order to answer questions that differ from those of quantitative research, the methodology must still follow basic ground rules and hold up under critical appraisal. It is said that ‘the strength of qualitative research lies in validity or closeness to the truth’ (Greenhalgh and Taylor, 1997, p.740).

Elliott *et al.* (1999) published their evolving guidelines for reviewing qualitative research, partly in an attempt to encourage better quality control in such studies. Their guidelines were not intended to stifle creativity but rather to ensure that qualitative studies provide meaningful and useful answers to the questions that motivated the research in the first place - what Mays and Pope (2000) refer to as ‘relevance’. Elliott *et al.* (1999) provided seven criteria that they felt were especially pertinent to qualitative research: (i) owning one’s perspective, (ii) situating the sample, (iii) providing credibility checks, (iv) grounding in examples, (v) accomplishing general versus specific research tasks, (vi) coherence and (vii) resonating with readers.

The first three of these points were addressed within the methodology and chapter. ‘Owning one’s perspective’ or ‘reflexivity’ is said to require a description of personal

and intellectual biases such that the author's orientations and personal anticipations can be understood, thus enhancing the credibility of the findings and allowing readers to interpret the results within this context (Mays and Pope, 2000). The researcher has also attempted to 'bracket the natural attitude' (Husserl, 1931) or set aside personal biases, assumptions and expectations as far as possible. Nevertheless, it is likely that personal presuppositions will have had some influence over the possibilities that have been allowed to emerge from the interview data.

In the current study, basic descriptive data was provided to describe or 'situate' the sample in as much detail as possible without compromising their confidentiality. This allows the reader to judge the range of persons and situations to which the findings might be relevant.

Credibility checks were used throughout the study and these are described in the method chapter. A clear exposition of methods of data collection and analysis was provided, with the grounded theory procedure in this study being explained and illustrated in some detail to allow appraisal of the fit between the data and the researcher's understanding of them. This was further supported with continual verification and suggestions for improvement being provided by two experienced clinical psychologists looking over the analysis and the supporting data to ensure the credibility of the categories, themes or accounts.

In the results chapter, the fourth criteria was adhered to throughout. Each theme was illustrated with examples from the data and elements in the data that appeared to

contradict the emerging explanation were provided. This type of presentation is sometimes known as 'fair dealing' (Mays and Pope, 2000, p.51) and helps to ensure that the viewpoint of one group is not presented as if it represents the sole truth about a situation.

The fifth criteria that Elliott *et al.* (1999) propose, requires researchers to appreciate the limitations of their interpretations given the range of informants and situations that have been studied and described. The current study undertook the research task of providing an understanding of the issues experienced by this particular group of parents of children with AIS with a focus on the issue of disclosure. This approach provides a *general* understanding and is based on an appropriate range of instances (i.e. 20 interviews). It is therefore likely to generalise to other parents with children with AIS within the age range of the current study.

Caution would need to be exercised, however, in generalising from the results of this study to other intersex conditions given the variation in physical appearance and medical difficulties involved in other diagnoses. Issues of gender were seen to be significant in the current study and may not apply so strongly in other intersex conditions where the sex of rearing may not be incongruent with the chromosomal sex. Even within AIS, there may be difficulties in applying the outcomes of the current research to older populations where the parents received the diagnosis many years ago. Medical attitudes and procedures have changed greatly over the years and the experiences for the parents in the current study, may be qualitatively different from that of parents whose children are much older.

Finally, coherence and resonance have been addressed throughout the study. The researcher has attempted to provide an understanding of the underlying structure of the analysis whilst preserving the nuances in the data. An integrated summary of each section of the analysis is provided in the results chapter to increase clarity and a verbal narrative is provided with each presentation being completed with a comprehensively explained and succinctly named 'central category'. The aim throughout has been to present the material in such a way that the reader's understanding and appreciation of the area might be significantly enhanced by an accurate and transparent representation of the subject matter.

Implications for Future Research

This initial study has provided a mass of rich data of which the key points have been considered above. Many ideas for future research are suggested by the limitations and outcomes of this study and some of these will now be discussed.

Parents' inability to take in the verbal information given at the time of diagnosis, plus a lack of printed information available to them to take away and consider later, may have major implications for their understanding and acceptance of AIS. The current study did not provide an insight into the actual information given to parents in the diagnostic session, nor did it directly assess parental coping per se. Further research might consider the information parents are given and the degree to which they are able to understand and retain this information. This could then be correlated with the

measure of parental adjustment and coping, with the hypothesis that less well informed individuals would have relatively greater difficulties in dealing with the diagnosis.

Parents expressed an opinion that they would benefit from meeting with others with children with AIS. Many felt that this would be more helpful than attending the current support group or meeting with a psychologist in the early years. The current study was unable to make a judgement about the relative value of these different forms of support. A research study that compared the type of support parents received with measures of their psychological adjustment and level of coping would be useful in guiding services in how they might best meet the needs of this group.

Parents' experience of the medical system is an area that deserves much further research. In the current study many parents commented negatively on their experience and it would be useful to know why this was the case. The interview data suggested that an individual's social class and educational level may affect the way in which doctors relate to them, however, the qualitative nature of the study allows only for subjective interpretation at this stage. Research which focuses on the quality of interaction between parents of children with AIS and their doctors, might highlight the presence and impact of any difficulties and ultimately help to foster better relationships between professionals and parents. Hopefully, through better engagement with services, parents would benefit more from the resources available to them and develop a better understanding of and adjustment to AIS so that they will be better placed to help their children learn about and accept their condition.

Parents in the current study commonly reported being afraid that the disclosure of AIS to their children would trigger significant psychological distress. There appears to be no literature looking at this aspect of the disclosure issue and the current study was only able to rely on one parental report measure (the CBCL) which looked for the presence of behavioural markers of psychological problems. Only four of children whose parents were interviewed had actually received a full disclosure of the condition and two of these girls did appear to experience significant problems. However, the data collected in the current study were not sufficient to allow conclusions to be drawn about any possible link between psychopathology and the receipt of a diagnosis of AIS. This is something which warrants further research to look for possible interactions between factors that might contribute to a child's ability to cope with the diagnosis. Such information might help professionals to suggest the most useful disclosure strategies to avoid such difficulties and to provide appropriate support to children and their families around the time of disclosure.

Questions of gender; the impact of the condition on gender role, gender identity and sexuality, were all raised within this research project. This qualitative study was not able to provide answers to such questions and there are a number of ways in which future research might prove useful. One such area concerns the extent to which parents' acceptance of their child's gender is related to the degree of virilisation at birth. The participants in the current study were parents of children with both CAIS and PAIS although, because of the relative prevalence of the two forms of the condition, there were many more parents of children with the complete form of the

syndrome. Whilst the majority of issues covered by the interview schedule appeared similar for parents of children with CAIS and PAIS, clearly this is one area where the two groups need to be separated and compared.

It would also be useful to discover to what extent parents' concerns about their child's gender identity and role are justified and to what extent they are related to the knowledge that their child has chromosomes which are generally described as being 'male' and, in some cases, were born with genitalia that were not fully female. At this stage, it would appear that parents of children with PAIS have more concerns and worries related to their child's gender identity and role. However, similarly to the above point, this hypothesis requires a much more detailed investigation. If the issues are different for parents of children with partial compared to complete androgen insensitivity syndrome, then those working with the families will need to be aware of these differences so that they can tailor their support accordingly.

In terms of the disclosure of AIS to the children, the study has shown that many parents may begin the process of disclosure later than is advised in the existing literature. However, the literature in this particular area is limited and appears to be based on the opinion of clinicians rather than being based in any specific research. It is not clear from the current study whether the relatively later age at which the children are given the first stage of the information has any negative impact on their well-being. It may be that this delay is helpful for the children or it may be that the parents simply put it off due to their own fears and thus deny the children the opportunity to learn about and come to terms with the information at a younger age.

This is an area that would benefit from further study which might inform both parents and professionals of the most appropriate disclosure pattern for children with this specific condition.

The parental experience of having a child with an intersex condition such as AIS is an area which has been researched only in a limited or indirect way until now. It was felt, therefore, that the 'discovery oriented' approach offered by grounded theory was the most appropriate way in which to investigate this area and the data collected have provided a detailed understanding of the meaning or nature of the experience for the participants (Stern, 1980).

From this detailed insight into parental experience, the study suggests a variety of areas that deserve additional research to begin to quantify the experiences and to compare across groups of participants. It is hoped that current study will inspire further investigations into a much under-researched area.

Clinical Implications

The current study gives rise to a number of implications for clinical services. Parents raised many issues within the interviews that should be addressed and it is hoped that through raising awareness of these, services might evolve to better meet these needs.

The time of receiving the diagnosis for their child is clearly of key significance for parents and the way in which this situation is handled appears to have a direct

bearing on the degree of understanding parents have about AIS and perhaps on their adjustment to and coping with the news. We know from the general literature on parental reaction to diagnosis, that receiving the news can pose a considerable threat to the larger representational system of caregiving. A lack of resolution can lead to parenting difficulties (Fraiberg *et al.*, 1983) and is strongly associated with insecure attachment between child and parent (Marvin and Pianta, 1996). The degree to which the person can mostly accept the permanence of the loss and reorient to the present and future is believed to reflect resolution of the trauma (Fonagy *et al.*, 1993).

In terms of implications for health care systems, it has been said that the process by which the diagnosis is delivered and information is fed back to parents can be a factor that, at best, does not facilitate resolution and in some cases militates against it (Pianta *et al.*, 1996, pp 254). A prolonged diagnostic process can be in itself traumatising to caregivers and the caregiving system, however, resolution will be enhanced if parents are provided with accurate information in understandable terms. They will thus be allowed the opportunity to integrate the difficult emotions and the changing perceptions of self and child that are part of this period (Pianta *et al.*, 1996).

Although the interviewees were divided as to whether they would have welcomed 'counselling' at this time, it is likely that there is a role for clinical psychology to liaise with the medical teams to ensure that parents receive a basic package of care when the diagnosis is made. It is suggested that in centres where this type of diagnosis is regularly made, that a protocol is established to ensure that parents gain the maximum benefit from the resources available.

This type of protocol would include; (i) printed information for parents giving a basic outline of the condition and its implications, (ii) information regarding sources of support for parents including clinical psychology services, medical specialists, parent support groups etc. and (iii) a follow up appointment within a month of the diagnosis being given. This type of appointment could perhaps be jointly provided by a clinical psychologist and clinical nurse specialist to address the immediate needs that parents have at this time and to establish links for the future.

Parents frequently reported that they had never had the opportunity to talk freely with someone who would 'really understand' their situation, i.e. another parent of a child with AIS. Other parents commented on their willingness to offer this type of support to others. It seems that both the need and the resource to provide this type of support are available and it might be useful for a support group to be set up, specifically for parents of children with AIS. This would enable doctors and other professionals to refer parents to this group where they might be put in touch with specific individuals who could talk to them at this time. It is not likely to be appropriate for services to take a lead role in setting up this type of support network since many of the issues that parents need to discuss are linked to their experiences of the medical system in particular. However, once established, the group might wish to call on outside agencies to provide information and support as required in a manner similar to the current support group.

The issue of the disempowerment of parents within the medical system is one that services clearly need to address. It is likely that the medical teams are unaware of the negative way in which their procedures and interactions are experienced by parents. For parents, and therefore their children, to gain the maximum benefit from the services within which they are involved, the professionals working with them need to adjust their style to improve the engagement with families. Clinical psychology could play a direct role in facilitating this type of training with the medical teams and to support them in changing their procedures and interactional style to enable to parents to feel more empowered and thus more able to cope with their child's confusing and sometimes distressing condition.

The need for research to investigate the incidence and prevalence of psychopathology in children following the disclosure of AIS has already been discussed. Parents clearly worry about this possibility and medical professionals need to be watchful of such signs and ready to refer to clinical psychology wherever appropriate - either to support children who are finding it difficult to cope with their diagnosis, or to work with parents on their fears and to enable them to find ways to adjust to the situation. If psychopathology is found to be a significant risk for these children, clinical psychology would then be advised and able to provide both preventative and reactive therapy to reduce the impact of such symptoms.

However, psychology services can only work at a preventative level if contact is made with parents in the early days and maintained (even if only on a six monthly basis) over the years. It is not clear how realistic such a proposal is, but by engaging

families with psychology services as 'a matter of course' when the diagnosis is made, parents may be enabled to access a greater degree of support at the times when it is needed most. This type of system would ensure that parents are linked in with a wider range of services and do not need to forge new relationships at times when there are difficult issues to deal with.

A major focus of this study has been around the issue of disclosure to a child of their AIS. Clearly parents experience significant difficulties in deciding what, when and how to tell their children about AIS and this is an area that needs to be given much thought by services. To be able to make the disclosure to their child, parents need support in a range of areas. They need to have received clear and accurate information and perhaps additional help to process and understand this fully. Once they are aware of the details of the condition and its implication, they will need to work through any concerns and fears they have about telling their child about AIS. They will then need to think about how they will impart the information to their child and the appropriate timing for this.

Many parents reported that although they were not sure about the idea of having 'counselling' regarding their child's diagnosis, they would particularly appreciate some help in thinking about the disclosure issue. Many were open to the idea of meeting with a clinical psychologist for help with this issue but it was rare for an interviewee to have actually been offered this opportunity. The disclosure issue raises a number of needs that could be met by services. There is a need for general information for parents which gives them an idea of the type of timescale for the

disclosure and also suggestions for how this might be achieved. In addition, it may be appropriate for services to adopt a protocol whereby all families meet with a clinical psychologist on at least one occasion to discuss this issue.

It may be that the reason parents appear to delay the disclosure is because of their uncertainty about how to go about this process and their fear that they might cause damage to their child by 'doing it wrong'. Sessions with a clinical psychologist could be used to help them think about their reasons for and against disclosure and to devise strategies that they can use to help them to tell their children about the condition. It may even be possible to do this type of work in a group setting, however, due to the relative rarity of the condition, it may not be feasible to gather a suitable number of parents together for this purpose.

Conclusion

The current study aimed to discover what the experience is for parents following the diagnosis of androgen insensitivity syndrome for their child and to understand better the issues that affect parents' decisions about whether, when, how and what to disclose to their children and others. A 'discovery oriented' approach was used to offer insight, enhance understanding and provide a meaningful guide to action (Strauss and Corbin, 1998).

The study describes in detail, for the first time, the experience for parents. It highlights a number of areas of unmet need and gives rise to many implications both for future research and for the development of services. Currently, parents' experiences often leave them feeling confused and alone and powerless to help their children. A more integrated model of care between medical and psychological teams is recommended so that parents might be offered better opportunities to absorb and adjust to the diagnosis, to make informed decisions about treatment and to appropriately disclose the details of the condition to their child.

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APPENDICES

Appendix 1 Letter to Participants

Name
Address

date

Dear

Re: Research Study

We are currently undertaking a study to look at the experience of parents when their child is diagnosed with an intersex condition such as androgen insensitivity syndrome (AIS).

Recent articles consider the question of whether or not children should be told about their intersex condition and the literature suggests that the medical profession are beginning to encourage greater openness about the diagnosis to the patient. However there is almost no published literature which takes into account parents' feelings about whether the intersex diagnosis should be disclosed to their child. We know little about the issues parents face when considering whether to tell their child, what to tell them and when.

We hope that this study will highlight the issues for parents and give a greater insight into their hopes, fears and needs. By enabling professionals to gain a deeper level of understanding of the issues parents face, they might be better placed to offer appropriate advice and support to families in the future.

I am therefore writing to ask if you might be interested in taking part in this study. Participating would involve you meeting with the researcher (Kate le Maréchal) and spending approximately 1½ hours taking part in a semi-structured interview. The questions will ask about your experiences related to your child's diagnosis of an intersex condition. In particular, the interview will be concerned with your thoughts about telling your child and others about their condition. We would hope to undertake these interviews at Great Ormond Street Hospital but it may be possible for the researcher to visit you at home if you are unable to travel to the hospital.

You do not have to take part in this research and a decision not to participate will not in any way prejudice any present or future treatment you or your child might receive. We are aware that taking part in research is time consuming, but your input will be invaluable and we would gratefully appreciate your contribution.

Only the researchers will know the identity of those who take part and any responses you give will be entirely confidential and anonymous. The researcher (Kate le Maréchal) will contact you shortly to confirm whether you might be willing to take part in this study. If you have any queries in the meantime, please do not hesitate to contact:

Kate le Maréchal (Clinical Psychologist in Training)

or

Polly Carmichael (Principal Clinical Psychologist and Supervisor) at:

The Department of Psychological Medicine
Great Ormond Street Hospital for Children NHS Trust
Great Ormond Street
London
WC1N 3JH

020 7829 8679

Yours sincerely

Kate le Maréchal
(Clinical Psychologist in Training)

Dr Polly Carmichael
(Principal Clinical Psychologist)

Appendix 2 Participant Information Sheet

Title

Telling Children About Their Intersex Condition: Issues for Parents

Aim of the study

The aim of the study is to look at the experience of parents when their child is diagnosed as having an intersex condition. In particular, we would like to find out more about parents' decisions about what and when to tell their child about the diagnosis.

Why is the study being done?

Although previous research has looked at the perspective of the individual growing up with an intersex condition, there has been very little that considers the experience of parents. It is hoped that this study will highlight the issues for parents and give a greater insight into their hopes, fears and needs so that families affected can be better supported by professionals involved in the care of the child.

There is a growing body of literature that looks at the question of whether or not children should be told about their intersex condition and at what age this should happen. However, when opinions are given they tend to be those of the medical professionals involved in the child's care. There are almost no articles which look at parents' opinions, the reasons for their decisions about what to tell their children and when. It is hoped that if clinicians are made more aware of the issues for parents, they will be better placed to offer appropriate support and advice.

How is the study to be done?

Parents of children with intersex conditions will meet with a Clinical Psychologist in Training working at Great Ormond Street Hospital to discuss their experiences of having a child with an intersex condition. The interview will last approximately 1½ hours and will be tape-recorded so that the researcher can make sure they have an accurate record of the material covered. The interview will cover areas such as your experience of receiving the initial diagnosis, different types of support that you may draw on and any thoughts you have about talking to your child about their condition.

Are there risks and discomforts?

There are no anticipated risks to this project. Confidentiality will be carefully maintained so that any material used will be fully anonymised and issues discussed in the interview will not prejudice further treatment in any way.

We are aware that talking about these complex issues can be difficult. There would be an opportunity for follow-up meetings, if participants desired, to discuss further any concerns that the content of the interview may raise.

What are the potential benefits?

The study may not bring any immediate benefit to you or your child. However, it is hoped that from the outcomes of the research, professionals will be able to gain a deeper level of understanding of the issues parents face. As a result, such professionals might be better placed to offer appropriate advice and support to families in the future, thus reducing the stress experienced.

Who will have access to the case / research records?

Only the researcher and her supervisors and a representative of the Research Ethics Committee will have access to the data collected during the study. Data will be anonymised with only the researcher being aware of the codes used.

Do I have to take part in this study?

No. If you decide now, or at any time during the study that you do not wish to participate in this research project, that is entirely your right. A decision not to participate will not in any way prejudice any present or future treatment you or your child might receive.

Who do I speak to if problems arise?

If you have any complaints about the way in which this research project has been or is being conducted, please, in the first instance, discuss them with the researcher, Kate le Maréchal (Clinical Psychologist in Training) or with Dr Polly Carmichael (Senior Clinical Psychologist). If the problems are not resolved or you wish to comment in any other way, please write to:

The Chairman of the Research Ethics Committee
The Research and Development Office
The Institute of Child Health
30 Guildford Street
London, WC1N 1EH

Or, if urgent, please ring the Committee Administrator on 020 7242 9789 (extension 2620) who will put you in contact with the Chairman.

Researchers who will have contact with the parents

Kate le Maréchal (Clinical Psychologist in Training) and Dr Polly Carmichael (Principal Clinical Psychologist and Supervisor).

Details of how to contact the researcher

You can contact Kate le Maréchal or Polly Carmichael at:

The Department of Psychological Medicine
Great Ormond Street Hospital for Children NHS Trust
Great Ormond Street
London, WC1N 3JH

Tel: 020 87829 8679

Appendix 3 Interview Schedule

(1) TIME OF DIAGNOSIS

- When was it first realised that there was a problem?

Prompts: *Age of child?*
 Memories of the way family/professionals reacted/managed the situation.
 What medical investigations were carried out? Feelings/concerns at the time?

- Can you remember when you were first given the diagnosis?

Prompts: *How long did you wait for the diagnosis after the problem was identified?*
 Who explained the diagnosis? Understanding of diagnosis?
 Memories of any discussion about sex assignment?
 Impact of diagnosis - i.e., did things change when a diagnosis was given?
 Feelings/concerns at the time?

- What support did you receive at the time of diagnosis?

Prompts: *Professional i.e. medical/psychological etc.*
 Family and friends

- What is your understanding of your child’s diagnosis?

Prompts: *Who has talked about this?*
 Has this been helpful/unhelpful?
 How else (if at all) have you found out about the diagnosis?
 Internet, support group, friends/family
 What terms have been used to describe your child’s condition (AIS, testicular
 feminisation, intersex)? How do you feel about these terms?

- Are there any medical/psychological implications of the diagnosis that you are aware of?

Prompts: *Current or long term?*
 Concerns in relation to these?

- What treatment has your child received?

Prompts: *What was that for?*
 Awareness of any other planned treatments/surgery in the future? (e.g. HRT, genital
 surgery)
 Awareness of duration or any long term effects of treatment(s)?
 Was there ever any discussion about sex assignment?

- How has your child been prepared for (told about) the treatment she receives?

Prompts: *What has been said to the child, who has said it?*
 Will this be the same different for any future treatments?

- What involvement have you had in treatment / management decisions that have been made?

*Prompts: Who makes decisions re: gender assignment / surgery / medical / psychological management?
Who do you think should make these decisions?
Satisfaction with way treatment and management decisions have been made? If not what needs to change to achieve this? i.e., understanding/consultation
What, if anything, would make the management by the medical team better?*

(2) People Involved / Support for the Family

- Which professionals have you had contact with in relation to your child's condition?

*Prompts: Hospital staff: psychosocial / medical /surgical /nursing
School - teacher / SENCo / Dr/nurse
GP practice- Dr/nurse/HV
How was contact initiated: pre-emptive or in response to a problem?*

- How have you felt about your contact with these people?

*Prompts: Adequate/helpful/unhelpful
Have you ever had additional people sitting in on consultations? How have you felt about this?
Have doctors generally seemed well informed about your child's condition and management strategy?
Have you ever had any difficulty getting test results?*

- Have you ever had any contact with the support group?

How have you felt about this? Have you found this useful?

(3) Disclosure - Talking about your Child's Diagnosis

- Who (if anyone) have you told about your child's condition?

*Prompts: Have you and your partner decided together about what to tell whom? Are you in agreement?
What informs/influences your decision?
Have your thoughts about sharing your child's diagnosis changed over time?
What, if any, concerns do you have about sharing your child's diagnosis with family, friends, professionals?
Is there anyone you would like to share your child's diagnosis with but haven't?*

- Has telling other's about your child's diagnosis ever presented any difficulties for you or your child?

- How have people reacted to the diagnosis?

Prompts: Positive and negative experiences?

- How do you deal with enquiries about your child's condition?

Prompts: Does anyone ask why your child attends hospital? What do you say?

- What discussions, if any, have you had with professionals about telling your child about their diagnosis?

*Prompts: Do you think the professionals involved in your child's care have a view about sharing your child's diagnosis with them?
If so what is it? Do you agree/disagree?
Have you had specific advice about this - who from? Was this helpful/unhelpful?
Have you been offered/would you like any, support in telling your child about their diagnosis?*

- Do you think your child should know about their diagnosis?

*Prompts: What are your reasons for this?
Is there any aspect of the diagnosis that you think your child should not know?
Is there any aspect of the diagnosis it is difficult to talk about?
Have you thought about how and when you might tell your child about their diagnosis?
Who do you think is responsible for telling your child about their diagnosis?*

- What, if anything, have you or others told your child?

*Prompts: What prompted you to give this information when you did?
What was this experience like for you and your child?
What was the impact, if any, on you/your child?
Is there more you would like to tell them?
Have you made any plans about how and when to give your child information?
If not - what stops you telling your child about their diagnosis? - (don't know how to do it - worry about how their child will react to the information, will say to others, how child will feel about parent)*

- How much do you think your child knows/understands about their diagnosis?

*Prompts: How do they know this?
Gradual/ planned process or not?
Has your child ever asked for information - when i.e., time of hospital visits?*

- Have you and hospital professionals talked without your child present?

*Prompts: Who instigated this? Have you been given the choice?
Do you feel able to ask for your child not to be present in consultations?
Have things been said in front of, or to your child, which have caused concern to you or your child?*

Perceived Impact of Diagnosis/Treatment - Present and Future

- Does your child's diagnosis affect your relationship with her in any way?

*Prompts: Nature of the diagnosis?
Impact of hospital visits?*

- Has your child's diagnosis had any impact on other family members?

*Prompts: Siblings/grandparents.
Diagnosis/hospital visits?*

- In what ways do you think your child's diagnosis might affect them in the future?

Prompts: Academic achievements, Job Prospects, Relationships, Fertility, Medication, Sexuality?

- Is there anything else about your experiences related to you child's diagnosis and management of this that you would like to add?

Appendix 4 Consent Form

Great Ormond Street Hospital for Children NHS Trust and Institute of
Child Health Research Ethics Committee

Consent Form for PARTICIPANTS in Research Studies

Title: Telling Children about their intersex condition: issues for parents.

NOTES FOR PARTICIPANTS

1. You have been asked to take part in some research. The person organising that study must explain the project to you before you agree to take part.
2. Please ask the researcher any questions you like about this project, before you decide whether to join in.
3. If you decide, now or at any other time, that you do not wish to be involved in the research project, just tell us and we will stop the research. If you are a patient your treatment will carry on as normal.
4. You will be given an information sheet which describes the research. This information is for you to keep and refer to at any time. *Please read it carefully.*
5. If you have any complaints about the research project, discuss them with the researcher. If the problems are not resolved, or you wish to comment in any other way, please contact the Chairman of the Research Ethics Committee, by post via The Research and Development Office, Institute of Child Health, 30 Guilford Street, London, WC1N 1EH or if urgent, by telephone on 020 7905 2620 and the committee administration will put you in contact with him.

CONSENT

I _____ agree that the Research Project named
above has been explained to me to my satisfaction, and I agree to take part in this study.
I have read both the notes written above and the Information Sheet about the project, and
understand what the research study involves.

SIGNED	DATE
-----	-----
SIGNED (Researcher)	DATE
-----	-----

Appendix 5 Ethical Approval 1 - Great Ormond Street Hospital

Institute of Child Health

and Great Ormond Street Hospital for Children NHS Trust
UNIVERSITY COLLEGE LONDON

25 September 2000

Dr P Carmichael
Department of Psychological Medicine
GOS

The
child
first
and
always

30 Guilford Street
London WC1N 1EH

Telephone: 0171 242 9789
Fax: 0171 831 0488

Dear Dr Carmichael,

99BS19 Telling Children about their intersex condition: issues for parents.

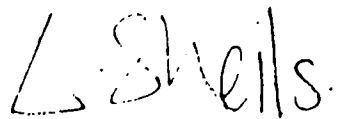
Notification of ethical approval

The above research has been given ethical approval after review by the Great Ormond Street Hospital for Children NHS Trust / Institute of Child Health Research Ethics Committee subject to the following conditions.

1. Your research must commence within twelve months of the date of this letter and ethical approval is given for a period of 12 months from the commencement of the project. If you wish to start the research more than twelve months from the date of this letter or extend the duration of your approval you should seek Chairman's approval.
2. You must seek Chairman's approval for proposed amendments to the research for which this approval has been given. Ethical approval is specific to this project and must not be treated as applicable to research of a similar nature, eg. using the same procedure(s) or medicinal product(s). Each research project is reviewed separately and if there are significant changes to the research protocol, for example in response to a grant giving body's requirements you should seek confirmation of continued ethical approval.
3. Researchers are reminded that REC approval does not imply approval by the GOS Trust. Researchers should confirm with the R&D office that all necessary permissions have been obtained before proceeding.

4. It is your responsibility to notify the Committee immediately of any information which would raise questions about the safety and continued conduct of the research.
5. On completion of the research, you must submit a report of your findings to the Research Ethics Committee.
6. Specific conditions pertaining to the approval of this project are:
 - The use of the enclosed standard consent forms for the research. A copy of the signed consent form must be placed in the patient's clinical records and a copy must be kept by you with the research records.

Yours sincerely



Orlagh Sheils
Secretary to the Research Ethics Committee

cc: Ms K le Maréchal, Clinical Psychologist in Training, GOS

Appendix 6 Ethical Approval 2 - The Middlesex Hospital



The University College London Hospitals
 The Joint UCL/UCLH Committees on the Ethics of Human Research

Committee A Chairman: Dr F D Thompson

Please address all correspondence to:

Iwona Nowicka
 Research & Development Directorate
 UCLH NHS Trust
 1st floor, Vezey Strong Wing
 112 Hampstead Road, LONDON NW1 2LT
 Tel. 020 7380 9579 Fax 020 7380 9937
 e-mail: iwona.nowicka@uclh.org

Dr P Carmichael
 Senior Clinical Psychologist
 Great Ormond Street Hospital for Children
 Department of Psychological Medicine
 Great Ormond Street
 London WC1N 3JH

November 21, 2000

Dear Dr Carmichael

Study No: 00/0263(*Please quote in any correspondence*)
 Title: **Telling children about their intersex condition: issues for parents**

The above application, previously approved by the Great Ormond Street Hospital, has been reviewed and agreed by Chairman's Action. There are no objections from the point of view of ethics. Please ensure that you have obtained final approval from the Trust (via the R&D office) before proceeding with your research.

Please note that it is important that you notify the Committee of any adverse events or changes (name of investigator etc) relating to this project. You should also notify the Committee on completion of the project, or indeed if the project is abandoned. **Please remember to quote the above number in any correspondence.**

Yours sincerely

p.
 Dr F D Thompson
 Chairman

Car21nv/ijn/November 21, 2000
 University College London Hospitals is an NHS Trust incorporating The Eastman Dental Hospital, The Hospital for Tropical Diseases, The Middlesex Hospital, The National Hospital for Neurology & Neurosurgery, The United Elizabeth Garrett Anderson Hospital and Hospital for Women, Soho, and University College Hospital.

Appendix 7 Demographic Questionnaire

1

Age of any children you have with AIS:

child 1

☐ years ☐ months

child 2

☐ years ☐ months

child 3

☐ years ☐ months

2

Do you have any other children?

If so, please give their age and gender

brother / sister 1

☐ years ☐ months

brother / sister 2

☐ years ☐ months

brother / sister 3

☐ years ☐ months

3

Children's ethnic group

4

Do any of your children have special needs (cognitive, physical, or educational)? If yes, please give details

5

Mother's age

☐ years ☐ months

6

Mother's occupation

7

Mother's ethnic group

8

Father's age

☐ years ☐ months

9

Father's occupation

10

Father's ethnic group

11

Are the parents ...

together? ☐ or separated? ☐

12

County where the child or children live at present (e.g. Essex, Berkshire, Kent, Sussex, Cambridgeshire etc.)

Thank you for completing this questionnaire.

Appendix 8 Complete Interview Transcript

Interview with Participant 7

Int: The place I'd like to start is when you first found out there was anything unusual or different about (*daughter*) - whether that was at the birth or later on.

P: No. It was ... erm ... she developed a hernia and went to the doctors and he ... erm ... booked her in for an operation ... erm ... but they couldn't do it 'til she was six months old so ... we waited until then, went along, went ahead with the operation and then they came out to me - this what at a local hospital in ... here. Err ... they came out to me and said that there'd been a ... they'd found something that they hadn't expected. So that was when we found out there was something wrong.

Int: So you'd been to your GP ... sometimes it's picked up that there's a hernia at birth.

P: No. No, it used to just pop up when she was really crying a lot.

Int: You went to your GP, your GP referred you on to your local hospital ... so what was that like when they came out and said 'We've found something'? What was your immediate thought?

P: I couldn't, I ... well when they told me what they found, I couldn't ... it didn't compute in my head. I couldn't understand what they were talking about because ... I mean, I think the words were actually used 'She should have been a boy' and I was ... I said 'What are you talking about?!', you know, because she physically looks so much like a girl ... erm ... that I ... I thought they had all gone mad! You know, I really thought they had all gone mad! What were they talking about?! She was so obviously a little girl. and ... erm ... it was just total shock. A complete shock.

Int: Yeah! Did they explain how they'd come to that conclusion?

P: Yeah, they ... well they told me that there were testes there and that she didn't have a womb, you know, and ... erm ... they gave me a ... they'd found a little slip of paper about AIS and they gave me the AIS support group thing and they gave me this piece of paper ... erm ... but ... and I gathered all my family around me and said 'Ooh there's something wrong with her, there's something wrong with her!' and

... erm ... made them listen to them saying what they were saying because I just couldn't ... couldn't believe what they were telling me.

Int: So that was when (*daughter*) was six months old?

P: Six months, yeah.

Int: So you had six months of everything was fine, apart from this hernia which popped up occasionally.

P: Yeah

Int: and would that have been a local paediatrician who came out to talk to you?

P: I don't know who it ... erm ... I think the surgeon came to speak to me ... erm ... and told me what they'd found and then they took me through to a consultant I think and I remember one of the things he said, 'cause I was obviously distraught - was crying and upset, and one of the things he said was ... erm ... that I will always remember, he said ... erm ... 'It's such a shame. We all expect our children to be perfect. My son was born with an incredible squint' and ... erm ... you know ...

Int: So he was comparing this to a squint?

P: Yeah. and I was like ... 'What?!' (*laughs*) 'What are you ... what are you on about!' You know, it was just ... I would be so pleased if she had a squint - if it was just a squint, you know.

Int: He was trying to say helpful ...

P: Yeah, I mean I suppose he was but to me, it was like 'What?!' You know, they're just ... they're telling me that this is freak. Basically. That's what I felt like. They're telling me I've given birth to something odd ... erm ... so it was really really weird. I felt sick; I actually went to the toilet and was benign sick - physically sick ... in the toilets when they told me.

Int: So actually receiving the diagnosis for you was a big shock - a horrendous experience.

P: It really was, yeah. It was awful.

Int: and you said you called in your family ... did you call your family and friends, did you say?

P: Well it ... erm ... (*daughter*)'s father and I had split up before she was born ... erm ... I called him and his family and my family, you know, like my mum, my sister. and so he came with his mother and ... erm ... yeah, they all heard the news at

the same time.

Int: and how was that, having them around? Was that helpful?

P: It was, yeah, I needed them there then. I needed them. I didn't need ... erm ... extended family then. I didn't want other people to know but I wanted them - I needed help to get through it.

Int: Right. So your immediate family and friends was important.

P: It was to me, yeah.

Int: So that was the moment of diagnosis. Did they offer you any kind of support at that time? Any kind of ... some kind of hospital based support - 'professional' support? Did anyone give you anything at all?

P: No, not really. They moved me ... they moved (*daughter*) from the main ward into a little room at the end of the ward ... erm ... and every ... I knew that everybody knew and I didn't like that. I didn't like the fact that all the nurses knew and people talked ... I felt like people were talking ... erm ...

Int: So you were moved into a room on your own at the end of the corridor - how did that feel?

P: It was better but again it was ... I mean it's difficult, I mean what can they do? You know, it was difficult. It was better because I felt people couldn't hear what we were saying and then, on the other hand, I was thinking they were all wondering what's wrong with her then. So it was ... it was all very strange.

Int: OK, so you didn't see a psychologist or a health visitor or ...?

P: No. No. The only thing they gave me was that slip of paper with the AIS support group.

Int: Did you contact them?

P: I did contact them, yeah.

Int: Was that helpful?

P: Yeah. That was ... I mean I contacted them after when we were home ... erm ... and yeah, it was, it was helpful.

Int: So you had a bit of information but not very much ...

P: No, not really.

Int: What happened next? Can you remember ...

P: Well then they ... then they talked to me about ... erm ... removing ...

because when they did the hernia operation they removed one testis for testing and then they talked to me about removal of the other one because they were saying to me about the ... erm ... chances of cancer developing if it was left and things like that.

Int: So did you feel quite involved in that decision?

P: Yeah ... err ... erm ... yeah, I was given the information and I - me being me - just wanted it out! (*laughs*) I just wanted them to do it, just get it gone immediately.

Int: What was that about?

P: Erm ... I didn't want there to be ... male ... male things there. Yeah.

Int: and how long did it take them to have the other testicle removed?

P: She was ... I think she was eighteen months old or just coming up to eighteen months old. I had to wait a long time and she went to (*National Hospital 1*) for that.

Int: So was that your first contact with (*National Hospital 1*)?

P: I can't remember if I went there before she had it done ... err ... I can't remember. I think she may have gone there before, just for some tests. We stayed overnight or something. But then we went back for the operation.

Int: and did you feel ... after you'd got the diagnosis, did you feel that you fully understood what the diagnosis meant? Did you feel that someone had explained it well enough to you so that you could understand what was going on?

P: I think I got more information from the AIS support group than I did from the hospitals really.

Int: So they'd given you the information 'she should have been a boy' and then it was left at that then?

P: See I can't really ... I mean I think I've blocked a lot of stuff out. I can't really remember but I ... I feel that I got more help from the support group than I did from medical people really.

Int: Sure. and can you remember ... it might be difficult to remember. Can you remember what your immediate thoughts were about the impact of this condition? What did you think it was going to mean ... for (*daughter*) or for yourself ...?

P: Erm ... well at first I was ... I didn't know what to think. I didn't know what to do. At first, to be honest, when I ... there was a point when I brought her

home and I was on my own with her, I didn't feel the same about her as I did before. Just a day or two. I went through a real weird, weird state ... err ... I just felt she wasn't my little girl that I was so proud of.

Int: Your little girl that you'd bonded with.

P: Yep. She was so beautiful and then they were telling me this and it was like 'What?' and I can remember one time she was in her cot and I was in my bed and I just felt like there was this strange thing (*laughs*) in the cot.

Int: Because everything you thought you were certain of had changed.

P: Yeah. Yeah. I can remember going through that but then that ... I got over that. Somehow, I don't know how. I just fell back in love with her I suppose. and felt protective towards her.

Int: and were you worried about things for the future?

P: Yeah, still am. (*laughs*)

Int: What were you worried about right back then? Can you remember?

P: Well the fact that she obviously won't be able to have any children ... erm ... I can remember ... I can remember asking doctors - this sounds really really weird 'cause she was just such a tiny baby - and I can remember asking them, 'Will she be able to ... erm ... have the sensations that a woman has? You know, will she be able to have sex? Will she be able to enjoy sex?' (*laughs*) You know, all these things and I can't even remember what their answers were but I can remember thinking things like that and thinking ...

Int: You were already thinking of her as a grown woman because of what you'd found out.

P: Yeah. and wondering what life would be like for her. The fact that she couldn't have any children - that's awful but a lot of women go through that and so I thought well hopefully she won't want any. You know, I just thought ... that was OK but the thought of her not being able to enjoy sex was like ... 'Oh god!', you know (*laughs*), can she not even enjoy that? You know, it was quite strange to be thinking of things like that for a tiny baby.

Int: and you do get thrown into the future don't you?

P: and now thinking about it, I didn't really get any information from them about ... erm ... the length of vagina and things like that. I didn't get any

information from them, from the doctors about that. I got that information from the support group.

Int: Right. So they were the people who were saying, 'Do you know what vaginal length she's got?' and 'Do you know ... have you thought about this?'

P: Mmm. Mmm. Yep. Because I still don't know. I still don't know that now. and I thought that was because perhaps she's not grown enough for them to know.

Int: Sure. Sure. OK. So you'd had a bit of explanation about the diagnosis, but not a lot.

P: Mmm.

Int: and you were worrying about ... about sexual relationships and about her having children. Were there other things you were worried about at that point?

P: Erm ... well would she develop like a normal woman?

Int: Yep. Do you mean physically?

P: Physically, yeah. Erm ... would she, would she ... erm ... be masculine? *(laughs)* Would she look feminine? You know? *(laughs)*

Int: Sure.

P: Feature wise, you know, as well. Things like that.

Int: and did any of those worries ... dissipate as *(daughter)* grew up or have they been things that have stayed with you?

P: Erm ... some of it stays with me ... erm ... because of the way *(daughter)* is. Erm ... she's very ... well that's *(daughter)* up there *(points to a photograph on the wall)*. She's ... erm ... very boyish! *(laughs)* She's very ... erm ... err ... she likes boys' things. She likes boys to play with. Erm ... she likes boys' toys, you know, things like that. and she says she would like ... she wishes she was a boy.

Int: Really?

P: So I think when that happens ... but I mean I don't know if that's all little girls ... or a lot of little girls go through that anyway, or ... but, but because I know what I know, that sort of comes up like a big red flag in my head whenever she says it.

Int: So if she didn't have AIS ...

P: If she didn't have AIS then I probably would think nothing of it.

Int: She's just a bit of a tomboy.

P: Yeah.

Int: Sure

P: But because she does and she says things, I think 'Oh no, please don't say that!' *(laughs)*

Int: Sure. It's a reminder ...

P: Yeah. and like yesterday, she did a valentine card and took a valentine card in for her little friend at school and I was so pleased! *(laughs)* I was thinking 'Oh good!' *(laughs)*

Int: *(laughs)* I'm not sure whether all parents would be pleased about that!

P: *(laughs)*

Int: So *(daughter)* had the hernia op. when she was six months and then she had the testes ... second testicle removed when she was eighteen months ...

P: Yeah I think she was about eighteen months old.

Int: Has she had any other surgery ...

P: No.

Int: ... or treatment?

P: No. She ... one thing I did ... when she went to *(National Hospital 1)* for the removal of the testes, they asked me could they take a piece of skin for DNA testing?

Int: Right.

P: and from that they would also be able to ... I'm really cross about this actually! They would be able to tell me if this just happened ... erm ... just a new thing that happened through me, spontaneously through me, or if I carry this and if it's carried through my family. and they told me that from that test they ... they would be able to tell me this. Now I've asked countless times for the results of that test and they've now told me - well last year they told me that ... erm ... no they didn't ... they didn't do anything with that.

Int: They took the skin sample ...

P: They took the skin sample and they've done whatever they've done with it but they ... they weren't able to tell me anything and didn't have any intention of telling me anything from that.

Int: and even when you phoned up to ask about it, they weren't able to tell you.

P: No.

Int: So that's left you feeling ...

P: It's gone to (*place 6*), I think they said ... erm ... and that I was told that that's what it was for.

Int: So they'd carried out a procedure ...

P: Yeah, they'd ... yeah. and she was very sore from that.

Int: Was that a vaginal skin ...

P: Yeah, just inside, yeah. They took a square piece of skin from her and she was very ... it was very sore and ... and, you know, if I had known that they wouldn't be able to tell me anything, or had no intention of telling me anything from that, I perhaps wouldn't have put her through that.

Int: No. Do you feel like ... you know, that she's had a painful procedure that you haven't benefited from?

P: Yeah and also, I ... from that I told my family - like my ... 'cause I thought well if ... if it is through like the mother's side of the family, my mother has six sisters and they all have daughters and it could happen ... it could happen! Again . to any one of those so ... something happened in the family and they were all gathered together and I told them. and I feel like I needn't have told them anything.

Int: Because you've no ...

P: Because I have no information to give them now.

Int: That's a difficult position to be in.

P: and now they all know about (*daughter*) and I wish perhaps they didn't

Int: Sure. So you told them because you felt you'd be able to give them some information that would be potentially helpful to them.

P: Mmm

Int: When actually you weren't able to give them that and you feel like ... is it like your trust has been betrayed?

P: Yeah. Yeah.

Int: So (*daughter*) hasn't had any more procedures other than that?

P: No.

Int: So when she was having her ... at eighteen months when she was having

the other testis removed, did you ... what did you tell people about why she was going into hospital?

P: Err ... I said that she'd ... erm ... I can't remember - what did I say?! ... I think I told them ... err ... I had one immediate friend I told. My family I told the truth and an immediate friend I told the truth. Erm ... I think I just said that she had another hernia and ... had to have ... I can't really remember.

Int: But it wasn't a big issue?

P: No, no.

Int: Sometimes people feel very anxious and very worried that everybody will know and they worry about trying to find a story.

P: I think I'd told people that she can't have children. That ... erm ... that she doesn't have a womb. But I haven't told them the full truth about ...

Int: Chromosomes ...

P: Yeah, I haven't told them that.

Int: and was (*daughter*) old enough to need explanation about it?

P: No. No, she doesn't even know that she ... well I've told her that she's had two operations 'cause she's got the two little scars there but she doesn't take any notice of them. She just ...

Int: It's just part of her.

P: Yeah. and there's photographs that I took of her in hospital and I said 'That's when you were in hospital' and she ... we went there when she was five and they measured her and weighed her ... erm ... and she didn't really know what it was all about. Just come along, you know, a day trip out to London.

Int: In Hamley's!

P: (*laughs*)

Int: OK. So she was fine about that?

P: Yeah, she's fine about that. and she knows that she can't have children. I told her that from day one, you know, that she can't have children.

Int: Sure. OK. Brilliant. Just checking that I've been asking you the things I'm supposed to ask! Does it feel OK so far?

P: Yeah, that's fine.

Int: I mean I think we've covered this a little bit but thinking about treatment

decisions, you said when they took out ... they took out the first testis to do tests and then they talked to you ... erm ... taking the second one out and you felt that you wanted that done.

P: Uh huh.

Int: Were there any other, sort of treatment decisions that have been made about ... I don't know ... for the future perhaps?

P: Talking about ... erm ... hormone treatments and I thought that ... the next visit is when she's nine. She's eight now. So I thought that's when they'd probably discuss that more. 'Cause I suppose she's got to be about eleven before that starts.

Int: Probably. and do you feel that you'll have a say in that? In deciding that?

P: I hope so, yeah. I mean I haven't ... I just go from one day to the ... you know, I just take ... I'm quite relaxed, quite a laid back person and I go as it comes, you know. I try not to worry about things too much and ... erm ... just handle them when they ... when they happen. Otherwise I'll just ... otherwise you just ...

Int: You go mad!

P: Yeah! (*laughs*)

Int: and just ... when (*daughter*) ... when you got the diagnosis - it might seem a strange questions - but was there ever any discussion about gender assignment? About whether (*daughter*) should be raised as a girl or ...?

P: No. Well ... they just ... there was no question really that she should be raised as a girl. They didn't ... they didn't say anything about her being raised as a boy.

Int: Sure. and you didn't have any questions about that?

P: No. No. Because to me she was a little girl so ... she is a girl so ...

Int: Sure. OK. That's fine. I'm just wondering if ... you said at the start that there wasn't really any support offered to you through the medical system. I just wondered whether you've has any contact with professionals since then?

P: I did. I went myself. I went to my ... 'cause I did go through depression and everything, but it wasn't just (*daughter*)'s condition. It was the break-up with her father and being on my own and everything ... erm ... because I was on my own until she was two and a half. So that period was ... erm ... I did go through a ... a depression stage and I went and saw a counsellor myself who had no knowledge of

AIS or anything like that so it was separate ... no it didn't ...

Int: So you were able, with your counsellor, to work through some of those things.

P: Yeah.

Int: Somebody outside of the immediate family.

P: That's right. and she wasn't ... also I think it helped that she didn't know anything about AIS or ... anything like that.

Int: She didn't have preconceived ideas.

P: Yeah.

Int: That was something you had to find for yourself?

P: Yeah.

Int: Did you go and ask your GP?

P: Yeah.

Int: Do you think that there's anything that would have been helpful to you, you know, if you'd been given carte blanche to decide, you know, I'd like to see this person and that person? What do you think would have been helpful?

P: I don't know. Maybe it would have been helpful if I had been offered ... erm ... counselling, but I don't think I was. Or if I was told that at a later stage, I could get some if I wanted some. But I was just in touch with the AIS support group ... erm ... and so I could talk to them if I wanted to.

Int: Did you go along to meetings?

P: I did go ... I did go to a couple but then ... err ... it was all ... it was OK but I found myself getting upset a lot of the time and I thought 'Well I don't need this really'. So ... I just ... left it alone. and there was ... the last ... there was a big one. I think it was the first big meeting they'd had where a lot of people got together and ... erm ... there were obviously parents that had just been told so were very sensitive and ... and ... erm ... I had (*daughter*) with me. She was only tiny and I'd said, you know 'Could some of the children meet?' and 'No, no, no, no, no!' They didn't want them to meet and I thought 'Oh, OK.' (*laughs*) So I didn't ... I didn't really go anymore because I thought it would be good if they could grow up with a friend that maybe had the same condition and they could maybe help each other through ... later on. If they developed friendships between themselves it might be good for them ...

you know, 'I'm not the only one that has this.'

Int: Sure. That's really interesting because some parents say, you know, 'I wish I'd been put in touch with just one other parent so I could know I wasn't the only one' and that would have been helpful. To think about it from the child's point of view - to be put in touch with another child ...

P: I just thought, you know, if it was somebody that they ... 'Oh yes!' ... you know, when it comes to the time of telling her, 'Well do you remember so and so?' that she has the same.

Int: Why do you think they were so against that?

P: I ... I suppose it was raw for them maybe. Maybe it was raw for them and perhaps they felt they were going through the, you know ... not wanting people to know. Or ... and I think one of them said 'Oh no, I don't ... she doesn't know you. She doesn't know who you are and they'll be wondering who you are and why you're there and ...' things like that. and I thought, 'OK. Fine.'

Int: Right. So for a support group they wanted to keep things quiet!

P: Maybe that ... that was just that particular family. I don't know. Maybe somebody else would feel differently. I don't know but ... I think most of the people were - the support group themselves - were people that had the condition and had grown or people that had grown up children ... you know, these particular people had a child around about the same kind of age and I thought that perhaps it would be nice if they could get to know each other but they didn't so ...

Int: That just goes to show how different peoples' experiences can be. The different stages that people are at.

P: Mmm

Int: So this is the bit that I sort of gave the title of my research to. About talking to children about their condition. and you'd said already that you've told (*daughter*) some bits. She knows that she won't have children and she knows that she's had a couple of operations when she was little. Have you ... I don't know what your thinking is in general about whether you want to tell (*daughter*) any more or ... whether you wish you'd told her less or ...

P: No. I don't wish I'd told her less. I do want to tell her. I do think it's important that she knows the full truth but I'm not sure when. I'm not sure how. I

don't feel now at all. I think she's ... she just wouldn't understand at all at the moment.

Int: Sure. Remind me how old she is?

P: She's eight. But she's a very young eight. *(laughs)* I don't know when the right time is going to be and that I'm hoping ... when I go ... erm ... to *(National Hospital 1)* ... I mean obviously I'm going to have to tell her something of why we're going next year. I don't know what I'm going to tell her yet ... erm ... I'm hoping then that maybe they'll be somebody there who can help me with all those ...

Int: There certainly is. I mean, for instance, *(Clinical Psychologist 2)* who I was talking about, is a permanent, full-time psychologist there. She often meets with families around that time ... to talk with parents and with children, if that's felt appropriate, about what's happening and how it can be explained and really thinking through ideas about that. So if you want to talk to somebody, she'd be a good person to talk to.

P: Yeah.

Int: and I think some of the doctors now ... it's much more in parallel between psychology and the medicine side of things now. Doctors tend to refer people on to the psychology side of things so that there's somebody to talk to about explaining those sorts of things so ... if that's something you want before you go down, you can always ask. So ... yeah. So thinking about, in advance about what to say and when to say it and how to say it ... they're very big questions.

P: Mmm.

Int: What does *(daughter)* understand so far?

P: She knows that ... erm ... you see the thing is she doesn't really know much about err

Int: Biology.

P: ... biology anyway, so ... but she knows that she hasn't got the equipment to grow a baby, I mean she saw me have ... grow pregnant ... have ... go through a pregnancy and everything and then was quite a good time to explain to her that ... erm ... she couldn't do that and I've told her about adoption ... erm ... when she was small and started playing with dolls - which she doesn't really do! and didn't really get into doing. But when we bought her dolls and things and then she'd go round

saying 'I'm going to have a baby when I grow up' and then I used to say to her 'Well, you know, maybe you will have a baby when you grow up but you won't be able to give birth to a baby. You won't be able to grow a baby inside you 'cause when you were growing inside mum's tummy, something went wrong and you didn't develop properly.' and that's all she ... that's what she understands ... but ... erm ... I haven't gone into ...

Int: How babies do grow!

P: No! (*laughs*) Erm ... so she knows that and she does understand and she did get a little bit upset about that and sometimes ... occasionally she'll come home from school and say something like 'I wish I could have a baby'.

Int: Right. So she has really taken it in.

P: Yeah, she really does understand and she knows about adoption.

Int: Right. So she knows that. and the rest of it - is there something that you think she should know, that you want her to know? Sometimes parents don't even feel that it's right that the child *should* know about their chromosomes or about ... erm ... the testosterone c^h ... the rest of the condition. Do you think she should know.

P: I think it's something she should know eventually, yeah. But not at the moment. She wouldn't understand.

Int: Sure. So it's a case of judging when she's old enough to understand that sort of thing.

P: Yeah. Yeah. I think ... I mean it's ... it's her. She's not me, is she? She's ... so yeah, she has the right to know that.

Int: and has ... has how much you've told (*daughter*), or not told (*daughter*) for now ... has that influenced what you've told other people?

P: My husband, obviously, gets quite cross with me if I tell anybody 'cause he says it's (*daughter*)'s business, not theirs and it's something that (*daughter*) should decide about who knows about her. Because I told his mum and he got annoyed with me for that. So it's difficult like when you're going to (*National Hospital 1*) like 'Well why are you going to (*National Hospital 1*)' you know. 'Why was she in there? What ...' It's difficult. It's hard to ...

Int: Sometimes it's easier just to tell them.

P: Just to tell the truth. But, I mean, I ... don't go ... my neighbours don't know. My good friend across the road, my neighbour across the road - she doesn't know. But ... I haven't told his brother - maybe his mother has, I don't know, but d'you know what I mean? It's like ... but (*husband*), he feels very strongly that it's (*daughter*)'s business.

Int: This is your current husband.

P: Yeah, that's my husband yeah.

Int: OK. and do you feel like that or do you feel less ...

P: Part of me does, yeah, part of me does feel like that and I think 'Oh I should have kept my mouth shut, I shouldn't have told that person.' At the time I had ... when I was going through it, I had a really close friend and I ... I told her but now we're not so close - not that we're not friends any more but we're not as close now as we were then and I think 'I wish I'd never told her that.'

Int: Yeah. When relationships change and you can't take back the information.

P: Yeah. That's right.

Int: I don't know whether you can even say, but do you know what it is that helps you make your decision about who to tell and who not to tell?

P: I think it is on how close I am to somebody and what's happening at the time, you know. Like I said it's ... his mum - I was going to (*National Hospital 1*) or we were going through photographs and 'Oh that was when she was ...' 'Well why was she in there?' It's hard to not just say.

Int: Sure. and what was her reaction? What's people in general's reaction when you've told them?

P: They feel sorry I suppose. They feel sorry and 'Oh what a shame' and ... I don't know. Probably that's about it really.

Int: and is that the sort of reaction that you want or ...?

P: Yeah. I don't want them to think 'Errrrrgh' (*expression of disgust*)

Int: Be repulsed by it.

P: Yeah, or feel ... or act differently with her.

Int: Yeah. Has that ever happened?

P: No. I don't think so.

Int: So they generally feel sorry that it's happened. They feel that it's a shame.

They don't ever react in a way that makes you wish you hadn't told them.

P: No. No.

Int: That's probably because you choose quite carefully the people who you tell.

P: Yeah. But then ... the reason I say 'Perhaps I shouldn't have told ... somebody' is because you don't know who that person then tells. It becomes gossip, doesn't it? So you think 'Oh, I wish I'd just kept my mouth shut' (*laughs*).

Int: Yeah. That's a difficult thing isn't it? It's not just deciding who to tell, it's deciding who they ...

P: and I think it's going to be the ... I think about that for (*daughter*) as well. That's another reason I don't know ... erm ... when she should know because children talk amongst themselves and I think ... she's not going to know ... the right person to tell or not to tell or ... you know and I think ... feel that perhaps ... erm ... she's got to reach a certain age where she's going to be able to recognise that. 'Cause I can't do it at my age! (*laughs*)

Int: So that's like two levels of development - one level where you're old enough to understand the diagnosis for yourself, however old that might be, and then another level where ... not only can you understand the diagnosis but you can understand who to tell and who not to tell and ...

P: Uh huh. Who to trust with information.

Int: So that's thinking for someone else isn't it? What will they do with the information? That's a hard level to reach.

P: It's like last week ... erm ... she's got a really good friend at school - her best friend at school, and ... erm ... she had to take in a toy. Now she's not allowed to take toys to school because she had something stolen but on the Friday, they had to take a toy in because they were doing this thing about limbs and the way limbs move. So she asked me, could she take this toy? and I said 'But you know you're not allowed to take toys in because of what happened before' and she said 'Yeah but I'll leave it with the teacher' and I said 'OK, you can take it in as long as you leave it with the teacher, you do not take it out at playtime, you don't leave it in your bag, you leave it with the teacher'. 'Yes mum, yes mum, I will'. I got to school to pick her up and her very good friend came running to me '*(daughter)* took her toy out at playtime!' you know, like this. and when (*daughter*) came up ...

(phone rings - tape paused)

and then when *(daughter)* came out, you know, part of me was really annoyed at that little girl, you know, instead of being annoyed at *(daughter)* for taking her toy out, I was ... I said 'Did you tell *(little girl's name)* not to tell me that you'd taken your toy out? 'Oh! Well everybody else was doing it mum and I really wanted to play with mine as well' and I said 'Yeah, well perhaps you ought to think about what you tell ... who you trust with these things', you know, 'cause I think ... and that was just a toy, you know.

Int: So something so important ...

P: Becomes gossip. Especially amongst teenage girls as well, I mean ...

Int: Oh yeah, of course.

P: It's going to be ... it's so difficult.

Int: 'Promise you won't tell' 'Yeah, I promise'

P: Yeah.

Int: and then everybody knows.

P: Yeah. So something's like that ... I feel like I ... in one way I don't want her to know so that she can't and then they can't all turn on her *(laughs)* d'you know?!

Int: Sure

P: and yet she does need to know.

Int: So it's giving her the responsibility - as well as the diagnosis you're giving her the responsibility for taking care of that for herself as well. Wow.

P: Mmm

Int: OK. This is a bit about the impact of the diagnosis and any treatment that *(daughter)*'s had. Do you think *(daughter)*'s condition has had any impact on your relationship with her, or your relationships as a family together or on your relationship with your husband? Do you think there's been an impact on relationships?

P: With her, yeah, I mean it all changed. It really did change when I was told but ... erm ... I don't know. I mean I don't know what it would have been like not knowing! *(laughs)* So I don't know really.

Int: of course. It is difficult. Sometimes ... one impact that people sometimes

talk about is their relationship with their partner. Sometimes people worry about ... feelings of guilt sometimes about whether the condition's been inherited, about who's responsible ...

P: Oh god, yeah! We went through all that yeah, yeah and I felt awful because it's through the mother ... so I ... I mean I carry guilt about that, yeah, sure.

Int: Was that something you worried about when you had your twins?

P: Oh god, yes! Oh that was awful! 'Cause I didn't intend to have any more children at all.

Int: and then you had two!

P: Yep. *(laughs)*

Int: Right. So you fell pregnant, was that immediately on your mind?

P: Oh god, yeah, yeah immediately and ... erm ... I actually went and ... well, not only ... I had to have it because one of them had ... erm ... a high risk of Down's Syndrome so I went and had the CBS and I was so ... I was scared because I thought I might, you know, I could lose one ... one or both of them, but I was glad because I knew that I would know what their chromosomes were. So ... erm ... yeah.

Int: and are they both boys?

P: They're both girls.

Int: They're both girls.

P: Yep. They're both girls.

Int: Without AIS?

P: Without AIS, yes.

Int: So you got the information that they were girls and so you knew, because they were XX ...

P: XX

Int: ... that they wouldn't have AIS.

P: Yep.

Int: Right.

P: But they could carry ... so I worry about that too.

Int: Sure. and you didn't get back the genetic information from *(National Hospital 1)* when you were supposed to.

P: No. *(laughs)*

Int: OK. I'm just wondering whether you ... again it might not be relevant but whether you think (*daughter*)'s diagnosis has had an impact on other family members.

P: I don't know about her father, I mean obviously he was involved in all of that but we never ... I mean we don't talk about anything really! (*laughs*) Apart from when are you going to pay me maintenance money! But ... erm ... I don't know about that side of the family at all but ... erm ... my mum, yeah, my mum was deeply affected by it ... erm ... my sister because my sister lost a daughter when she was eight so ... she immediately was thinking 'Oh, could she have had that?'. and ... erm ... my nephew has just had a little girl and they came and said 'Should we say anything about this?' and I said 'No you don't need to because, you know, he's male so ... he doesn't carry it or anything. It's got nothing to do with the fact ...'. I had to go through it all again and explain.

Int: So you're taking on the role of a geneticist and a paediatrician and a ... psychologist all at the same time.

P: Yeah.

Int: Does (*daughter*) see her biological father?

P: Yeah she sees him every fortnight, yeah.

Int: Do you think it's had any impact on their relationship? It's difficult to say ...

P: I really don't know. I don't know.

Int: I just wonder whether ... you said before that there were things that still worry you now for the future for (*daughter*). I wonder what the things are now that are foremost in your mind? You said when she was little you were thinking about the fact that she wouldn't be able to have children and ... sort of worries about sexual intercourse ... is it the same things now or do other things worry you?

P: I worry now about how she will react to the information when she's given it.

Int: What are your thoughts about that?

P: She is a very dramatic little girl. She ... erm ... she feels things very deeply and I'm ... I do worry about her ... how she'll take the news and respond to it.

Int: What are your worries ...

P: My biggest fear - that she will go so far into depression she will commit suicide! *(laughs)* That's my biggest fear. Worried that she won't form relationships with people.

Int: Would that be because she won't want to or because she ... won't be able to or ...?

P: I'm worry that she'll think she's strange and ... will go into herself ... erm ... because she's very much ... she's a very friendly little girl. She's so ... I mean my other two are so different to her, like she's really outgoing and really ... erm ... she'll make friend with somebody who's just come into the street for a look round, you know, she's very very like that and I'm worried it's going to make her go into herself and be quiet and shy and ... ashamed.

Int: Do you think that's because you think about how much of a shock it was to you when you found out?

P: Possibly. Possibly, yeah.

Int: Do you think there might be any positive aspects to her knowing?

P: Erm ... I don't know, I just ... because I ... I went to that meeting and there were women that had the condition and the truth was kept from them ... and they talked about their experiences and how they felt ... erm ... it wasn't fair that they weren't told the truth. That has stuck in my mind and I just ... I just feel it's important that she knows the full truth ... and that I'm not keeping anything from her. You know, it's a similar feeling to how I feel about her father, you know, 'cause I went through a lot with him and a lot of women in my position would have just blanked him out the door and gone and stay away and the rest of it. But I always felt that I don't want her to say to me 'Why didn't you let me see my dad?' and so I don't want her to say to me 'Why didn't you tell me the truth?'. You know? *(laughs)*

Int: So what you'd want for you, isn't necessarily the same as what you'd want for her. You'd put your own feelings aside ... that's very hard.

P: Well yeah, I mean those women that weren't told the truth feel ... felt cheated and felt that everyone else knew and they didn't and I think that's wrong. So ... I mean I do want her to know but I wish she could ... I wish somehow she could grow up *(laughs)* and I could tell her it all and then she could go back *(laughs)*.

Int: Yeah, so she's old enough to understand it all without it ... yeah.

P: Yeah.

Int: Do you have a sort of idea in mind of what sort of age you want her to know by?

P: No. No, I don't. I know that like when ... when the other girls are starting to develop and have periods and things, she's going to have to know something then. She's going to have questions so she's going to have to be answered but I don't know, I really don't know.

Int: It's a big unknown at the moment.

P: Yeah. It's difficult.

Int: But ... I mean you said you're going up to (*National Hospital 1*) next year so that's sort of a milestone in a way anyway ... thinking about that.

P: Yeah. See and I ... I have no idea about hormone treatment or anything. I haven't read anything, I haven't listened to anybody else's experiences, because I've found with the support group that they were telling me things and I was getting so confused that that's another reason why I blocked ... I didn't go anymore, although I have contacted them through the internet and I know that I can, so I know when the time comes I know that they're there. But I haven't subscribed to them and I don't want their newsletters and all their bits of medical information that comes piling through your letterbox ...

Int: It's a lot to take in.

P: It just ... I've got a file full of it in there and I keep thinking 'I'll throw that away', you know, 'cause she's going to find it and want, you know, 'Mum, what's this?'. You know, and ... erm ...

Int: I suppose when it's so complex, if it's complicated for you to understand and make sense of, then trying to think about telling (*daughter*) is even more difficult.

P: Mmm. Yeah. It just ... maybe it's selfish really but I just feel like we need to live our lives and when these things happen ... when these problems occur, then we'll deal with them then, you know, 'cause ... I keep getting all these things through the door and I keep seeing it every day and it just fills up your life and you just ... god ... and you've got two other children to deal with and ...

Int: Busy life!

P: and she's happy. You know ...

Int: OK. Well I've covered most of the things I wanted to ask you about. I just wondered if there's anything that you thought I might ask about or you'd thought would be important to talk about or anything that, in your experience, has been a really key issue that we haven't covered?

P: No, not really. I mean I ... like I say, I didn't think about you coming and what you were going to say or anything. I just thought, oh you'll come and I'll just say whatever I have to say and you'll say whatever you have to say (*laughs*). No. I think ... no, there's nothing really.

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